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Case Report

Unilateral nevus comedonicus associated with epidermoid cyst

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ABSTRACT

Nevus comedonicus (NC) is a type of an epidermal nevus which was first described in 1895 by Kofmann. Clinically, it is characterized by linear lesion comprising numerous dilated follicular openings with keratinous plugs resembling classical comedones, seen mainly on the head and neck area followed by trunk and upper arm. Treatment options of NC include topical therapy, laser, and surgery. NC associated with epidermoid cyst is rare. We report here a case of NC affecting left side of trunk, left lower limb with epidermoid cysts.

Keywords: Comedones, Epidermoid cyst, Nevus comedonicus

INTRODUCTION

Nevus comedonicus (NC) is a type of an epidermal nevus which was first described in 1895 by Kofmann.[1] Clinically, it was characterized by numerous dilated follicular openings with keratinous plugs resembling classical comedones seen mainly on the head and neck area followed by trunk and upper arm. It may be present at birth or develop throughout adult life. The prevalence of NC has been estimated from 1 in 45,000 to 1 in 100,000, with no gender or racial preference.[2,3] Epidermoid cyst, on the other hand, was common and most frequently presents in young and middle-aged persons. It results either from inflammation of pilosebaceous follicles or from deep implantation of a fragment of epidermis by a blunt penetrating injury or due to developmental defect. NC associated with epidermoid cyst is rare.[4-7] We report here a case of NC involving unilaterally on left side of trunk and left lower limb with epidermoid cysts.

CASE REPORT

A 11 years old female child presented with comedo-like lesions over the left side of trunk, left lower limb along the lines of Blaschko since birth which gradually increased in size and number. In due course of time she developed nodular swellings scattered within the area of these come done-like lesions for the last 2-3 years. The lesions were asymptomatic in nature. There was no family history of similar complaints. Cutaneous examination revealed groups of dilated follicular openings filled with keratin distributed over the left side of body with multiple nodulocystic swellings of the size 2-4 cm in diameter with scarring and fibrous tracts scattered in between. General, physical and systemic examination was normal. So, negative EEG abnormality, ipsilateral cataract, corneal changes, skeletal abnormalities were present. Routine laboratory investigations including complete blood count, blood chemistry, and urinalysis were within normal limits. Diagnosis of NC was made on the basis of history and clinical examination.

The histopathology of the comedo-like lesions revealed dilated hair follicles containing keratinous debris, consistent with NC. The interfollicular epidermis was normal. Sections from nodulocystic swelling showed cyst wall lined by stratified squamous epithelium with granular layer and lamellated keratin, consistent with

epidermoid cyst. Accordingly, a diagnosis of NC with epidermoid cysts was rendered.

Figure 1: NC in a linear pattern on left lower limb.

Figure 2: NC associated with epidremoid cysts on left groin and thigh.

Figure 3: NC on left side of trunk.

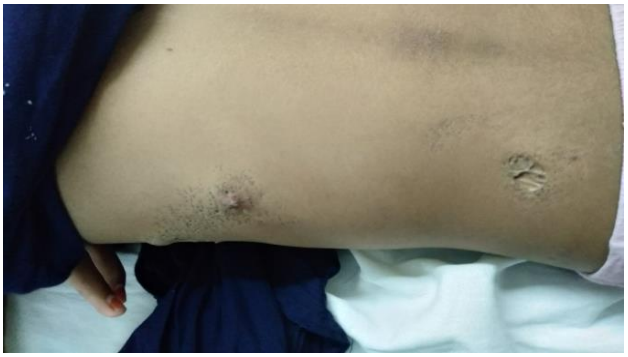

Figure 4: NC on left back.

Figure 5: NC on left leg in a linear pattern.

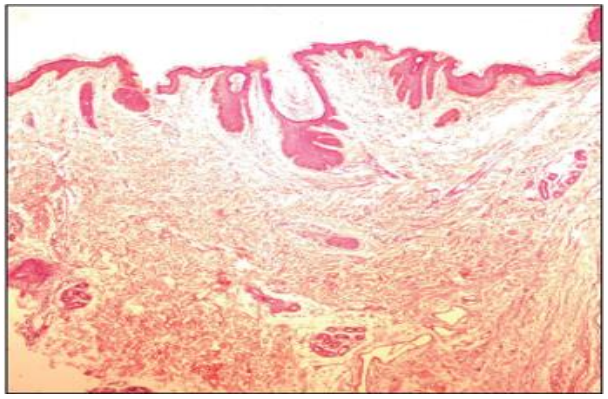

Figure 6: The histopathology of the comedo like lesions revealed dilated hair follicles containing keratinous debris.

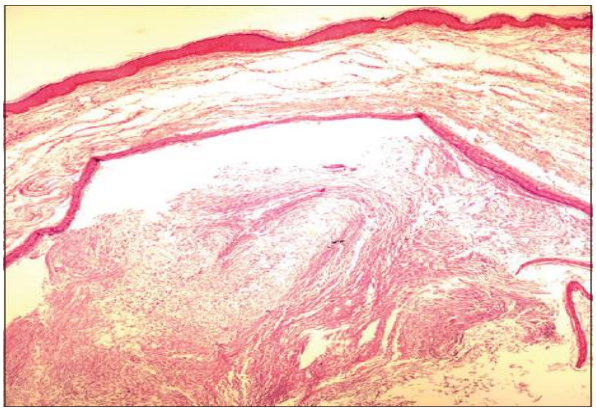

Figure 7: The histopathology of epidermoid cyst showed cyst wall lined by stratified squamous epithelium with granular layer and lamellated keratin.

DISCUSSION

NC can present in two clinical entities. The first is the asymptomatic comedo-like lesions and the second is the presence of cysts, scars, fistulas, and abscesses due to inflammation and infection of the comedo-like lesions.

The case described here is of the second group. NC may be linear, interrupted, unilateral, bilateral, along the lines of Blaschko or segmental. The characteristic histological feature of NC is an aggregation of deep, wide invaginations of acanthotic epidermis, filled with concentric lamellae of keratin.[2] There have been occasional reports of associated developmental anomalies including ipsilateral cataract, epidermal nevus, and trichilemmal cyst.[8-10] NC syndrome belongs to the large category of epidermal nevus syndromes in which epidermal nevus is associated with abnormalities of other body organs such as neurological deficits (epilepsy and electrocardiogram abnormalities), skeletal abnormalities (scoliosis and spina bifida), and eye problems (congenital cataracts). The present case has epidermoid cysts along the distribution of NC, which is a rare association.

Conservative management options include extraction of come done, and the use of topical agents such as retinoic acid, urea, tretinoin, ammonium lactate lotion, tacalcitol, tazarotene, and calcipotriene.[11-13] Conservative treatment may improve cosmesis in some patients but cannot prevent recurrence. Erbium-YAG laser and carbon dioxide lasers have also been reported to be successful.[14,15] Surgical excision is the treatment of choice for epidermoid cysts.

CONCLUSION

Here we report a case of nevus comedonicus associated with epidermoid cyst distributed unilaterally on trunk and leg in a linear pattern along the lines of Blaschko on left side. Localized nevus comedonicus can be removed by surgical excision followed by primary suture with good esthetic results but in this case as large surface area is involved it requires staged excision or skin grafting.

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