

Bilateral Naevus Comedonicus

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Abstract

Observations: Nevus comedonicus is a rare adnexal hamartoma of the pilosebaceous units. Here, a 20-year old man with bilateral nevus comedonicus of neck is presented. His nevus comedonicus, comprising of a linear group of hyperpigmented comedone like papules is seen on both sides of his neck. There was no associated pain, itching or discharge. He presented mainly for cosmetic reason. Skin biopsy of the lesion showed the presence of widely dilated invagination resembling dilated hair follicle filled with orthokeratotic horny materials penetrating the reticular dermis. The interfollicular epidermis was normal.

Introduction

Nevus comedonicus (NC) manifests as asymptomatic groups of closely set, dilated follicular openings filled with dark keratin plugs resembling comedones. The intervening epidermis may be normal. The majority of cases are isolated. However, NC may be part of syndrome in association with skeletal, CNS, ocular or cutaneous abnormalities called naevus epidermal syndrome [1, 2]. Although usually present at birth they can present later in life. The most commonly affected site is face, neck, trunk, upper arms, but scalp, palms, soles and glans penis may occasionally be involved. The follicular structure is not able to form matrix cells or hairs and is capable of forming only soft keratin which plugs the adnexal orifice and produces the comedones [3]. The etiology of NC is unclear. Why some NC patients present late in life is not known, although a genetic mosaicism has been proposed. While the majority of cases are sporadic, several families with this condition have been documented. Most patients

are asymptomatic. Uncommonly, the lesions become repeatedly inflamed and infected, leading to painful cysts, abscesses, fistula formation, and scarring [4]. Additionally, patients may be distressed over the cosmetic appearance of the lesions.

Case Report

20-yr-old man presented with asymptomatic grouped comedone like keratin filled pits on right side of neck since the age of 5 years. In due course of time similar lesions developed on left side with pubertal accentuation. There was no history of similar complaints in the family. Examination revealed multiple groups of dilated pilosebaceous orifices filled with black keratin plugs over both sides of neck (Figure 1). There were no CNS, skeletal, eye or other cutaneous abnormalities. Histopathology revealed rudimentary hair follicles and sebaceous gland with epidermolytic hyperkeratosis, findings suggestive of nevus comedonicus. No other laboratory test was done as no systemic abnormality



Figure 1. Bilateral naevus comedonicus in a linear pattern.

was detected clinically. Topical retinoic acid (0.025%) was given as treatment.

Discussion

Nevus comedonicus is an uncommon developmental anomaly also known as nevus acneiformis, zoniform nevus with comedones and systematized sebaceous gland nevus [5]. Many consider it as a type of adnexal hamartoma, with abnormal differentiation of the epithelial portion. The follicular structure that results is unable to form terminal hair or sebaceous glands and is capable of producing only soft keratin [3]. Others consider this lesion to be an epidermal nevus involving the hair follicle or an appendageal nevus of sweat ducts [6]. Nevus comedonicus comprises of groups of pits filled with black keratinous plugs resembling black heads. The intervening epidermis may appear normal, hyperkeratotic, hypo or hyperpigmented [4, 6]. There may be one or several lesions in a linear, unilateral [3, 7, 8] or rarely bilateral distribution [9]. Lesions may be very extensive [3, 8]. The commonest site is face followed by the neck, trunk and upper arm. Palms, soles and glans penis may be involved. The lesion usually manifests at birth or by the age of 15 years or rarely as late as middle age with no particular sex predilection. [3]

Clinically, nevus comedonicus can present in 2 clinical entities. The first is the asymptomatic comedo like eruptions usually seen on the face, neck, trunk and upper arm and the second is the presence of cysts, scars, fistulas, and abscesses due to inflammation and in-

fection of the comedo like lesions [4]. Albeit rare, there are also reported on the palms, scalp, ears and genitalia [10, 11, 12, 13]. Most of the lesions are unilateral and segmental, following the lines of Blaschko [14]. Association with extracutaneous manifestations especially neurological, ocular and skeletal anomalies have been termed nevus comedonicus syndrome [1, 2]. These anomalies include electroencephalographic abnormalities, ipsilateral cataract, corneal changes, hemivertebrae, scoliosis and absence of the fifth ray of a hand [15]. These anomalies include electroencephalographic abnormalities, ipsilateral cataract, corneal changes, hemivertebrae, scoliosis and absence of the fifth ray of a hand [15].

The various topical preparations used for the treatment are retinoic acid, 12% ammonium lactate, tazarotene and calcipotriene [3, 16, 17]. Surgical excision is more effective in the long term than superficial shaving or dermabrasion [3, 18, 19]. Our patient did not report for follow-up.

Our patient is a case of bilateral nevus comedonicus and we report this case for its rarity.

Conclusion

Nevus comedonicus is usually present at birth in a unilateral fashion, but in our case lesions appeared on neck at the age of 5 years with bilateral involvement which is very rare.

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