

## CASE REPORT

## A Case of Becker's Nevus with Osteoma Cutis

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Becker's nevus is a relatively common acquired focal epidermal melanotic hypermelanosis usually found in the region of the shoulder girdle of young men. Various skeletal malformations and cutaneous dermatoses have been reported in Becker's nevus. Osteoma cutis is a rare disorder characterized by compact bone formation in the dermis or subcutaneous tissue. Secondary ossification on nevi has been reported, but not with Becker's nevus. Herein, we report a case of Becker's nevus with accompanying osteoma cutis in an 18-year-old female. (*Ann Dermatol* 23(S2) S247 ~ S249, 2011)

**-Keywords-**

Bone neoplasm, Nevus, Osteoma, Pigmented

### INTRODUCTION

In 1949, Becker<sup>1</sup> described a hyperpigmented patch with hypertrichosis on the shoulder of two young men. This was later identified as an independent disease and named "Becker's nevus." Becker's nevus has unilateral hyperpigmented hairy patches, occasionally showing a bilateral distribution and having a high prevalence in Asian people<sup>2</sup>. Becker's nevus can be associated with various anomalies including skeletal or soft tissue defects, unilat-

eral breast or nipple hypoplasia and smooth muscle hypoplasia<sup>3</sup>.

On the other hand, osteoma cutis refers to new bone formation on dermis or subcutaneous tissue without any family history of Albright's hereditary osteoma cutis<sup>4</sup>. Osteoma cutis can be categorized by several methods but is usually divided by the presence of underlying diseases including skin tumors, associated trauma, or metaplastic ossification associated with neoplasm<sup>5</sup>. Dermatoses accompanied by osteoma cutis are trichoepithelioma, basal cell carcinoma, nevus, hemangioma, pyogenic granuloma, chondroid syringoma and dermatofibroma<sup>5</sup>. Becker's nevus with accompanying osteoma cutis has not previously been reported. Herein we report a rare and interesting case with a review of the literature.

### CASE REPORT

An 18-year-old female visited our outpatient clinic, complaining of a pigmented skin lesion on the right upper arm. The patient had been in her usual state of good



**Fig. 1.** Localized light-brown colored reticular patches with hair on the right upper arm.

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health, and had no remarkable abnormalities other than the skin lesion. The skin lesion had been there since birth and she had undergone several laser treatments before visiting our clinic. She had no remarkable family history of disease.

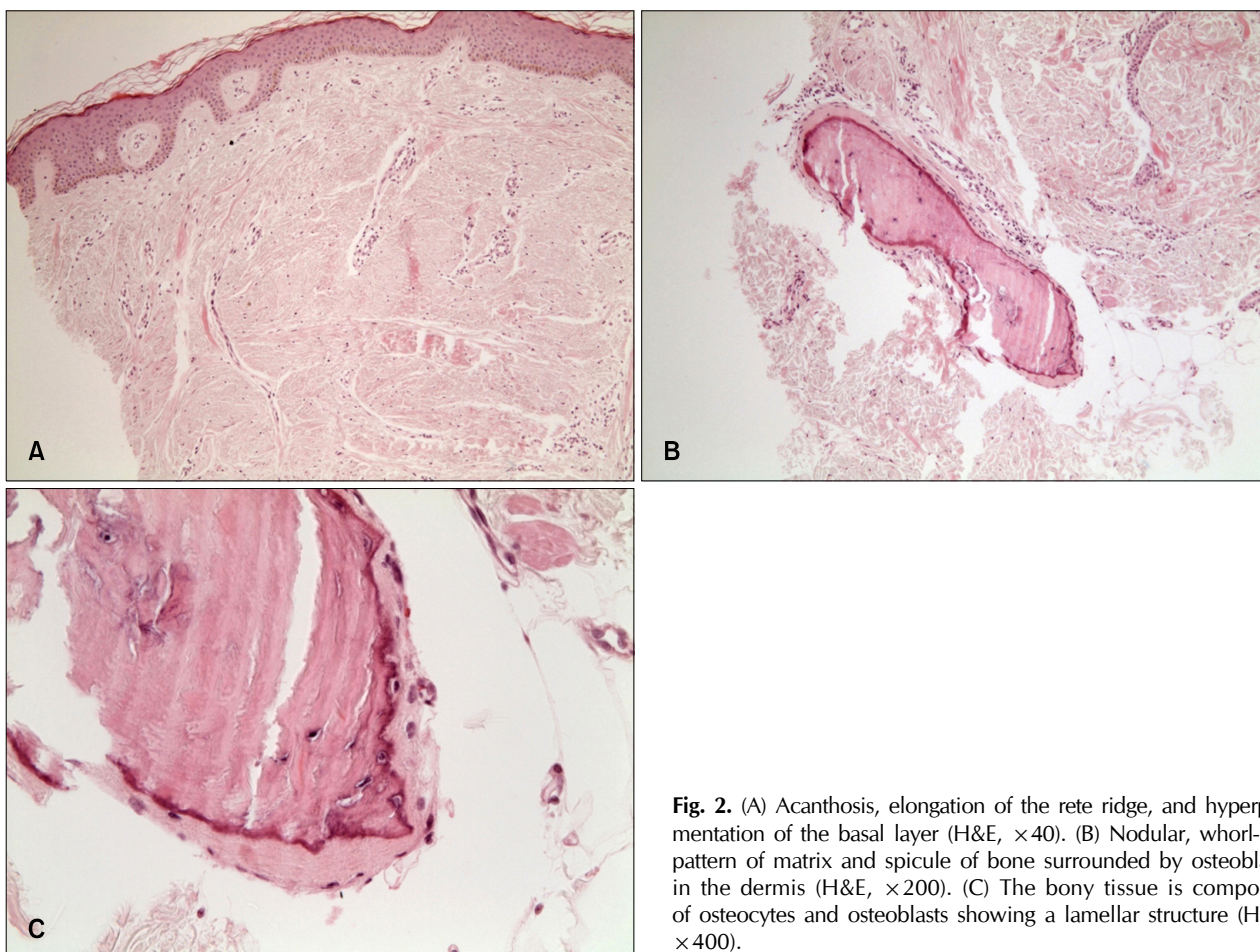
Skin examination revealed a well-defined, 10×8 cm hairy pigmented patch on the right upper arm (Fig. 1). The skin biopsy from her right upper arm showed hyperkeratosis of the epidermis, acanthosis, elongation of rete ridges and increased basal melanin pigmentation (Fig. 2A). The upper dermis showed a nodular, whorl-like pattern of matrix without nevus cells. There was an oval shaped compact bone formation on the lower dermis (Fig. 2B). Higher magnification revealed that the bony spicule had a lamellar structure with peripheral palisading of osteoblasts (Fig. 2C). The clinical appearance and the histopathological evidence were compatible with the diagnosis of Becker's nevus associated with osteoma cutis. However, since neither additional symptoms nor a palpable mass on the skin have been observed, the osteoma in biopsies is thought to be an incidental finding that requires no further

treatment. We gave the patient twelve sessions of ND:YAG laser treatment, which resulted in some improvement (reduction) in the pigmentation, after which the patient wanted no more treatment.

## DISCUSSION

Becker's nevus has also been called pigmented hairy epidermal nevus and occurs most commonly on the shoulder, back or chest of male adults, but may involve any area of the body. Androgen is thought to play an important role in the pathogenesis, as the lesion occurs after puberty and the condition has a higher prevalence in males<sup>6</sup>. Formigon et al.<sup>7</sup> described an increased level of androgen receptors on lesional tissue in a patient with Becker's nevus with ipsilateral breast hypoplasia, which he suggested as a cause for the associated abnormality.

The genetic evidence for this disease is not clear, but loss of heterozygosity of alleles is suggested<sup>8</sup>. Becker's nevus occurs in a sporadic fashion but sometimes occurs as clusters within a family. This can be explained by an early



**Fig. 2.** (A) Acanthosis, elongation of the rete ridge, and hyperpigmentation of the basal layer (H&E, ×40). (B) Nodular, whorl-like pattern of matrix and spicule of bone surrounded by osteoblasts in the dermis (H&E, ×200). (C) The bony tissue is composed of osteocytes and osteoblasts showing a lamellar structure (H&E, ×400).

postzygotic mutation in early embryos giving rise to mosaic clusters. As this phenotype would be neither simply mendelian nor entirely nonmendelian, Happle<sup>9</sup> proposed the new term "paradominant inheritance".

Glinick et al.<sup>3</sup> divided abnormalities into hamartomatous and hyperplastic or structural abnormalities. Happle and Koopman<sup>8</sup> suggested calling developmental abnormalities associated with Becker's nevus by the name "Becker's nevus syndrome". Possible structural abnormalities accompanied by Becker's nevus are underdeveloped breasts and developmental abnormalities such as limb hypoplasia, focal fat hypoplasia, scoliosis, intervertebral herniation and craniofacial abnormality<sup>10</sup>. There is no optimal choice of treatment for Becker's nevus, but there are several options in laser treatments, including ablative and Q-switched techniques<sup>11</sup>.

Cutaneous ossification can be classified into a primary form, where there is an absence of a pre-existing or associated lesion, and a secondary type, where ossification develops in association with a pre-existing skin lesion. Secondary ossification accounts for the great majority of cutaneous ossifications. Underlying cutaneous disorders of ossifications are nevi, basal cell carcinomas, pilomatricomas, hemangiomas, pyogenic granulomas, schwannomas, lipomas and chondroid syringomas<sup>5,12</sup>. Bone formation can also be found at infection or trauma sites such as acne scars, injection sites, hematoma and surgical sites. Abdominal wounds are particularly involved and it seems that injury to the xiphoid process or pubis may liberate bone-forming cells into the wound site. Other circumstances include chronic venous insufficiency of the legs, scrotal calcinosis, scleroderma and morphea<sup>5</sup>.

Burgdorf and Nasemann<sup>13</sup> explained that bone formation in the wrong site, first as misplacement of primitive mesenchymal cells of osteocyte differentiation potency, and second as metaplasia of under-differentiated or differentiated somatotic cells. The latter explanation seems more reasonable in secondary ossification.

Histopathologically, cutaneous ossification can be found in variable sizes in the dermis or subcutaneous tissue. It is composed of numerous osteocytes, cement lines and, occasionally, osteoclasts, fat cells and hematopoietic stem cells<sup>4,5</sup>.

In this particular case, the patient had a history of laser treatment on the site of the pigmented patch, which could have contributed to metaplasia of the osteocyte. To our knowledge, Becker's nevus with accompanying osteoma

cutis is not easy to find in the literature. It is not known if it is a hamartoma of Becker's nevus, a secondary ossification due to laser stimulation, or an incidental osteoma cutis on the under skin of the nevus. Repeated laser stimulation could have functioned as a physical stimulant for mesenchymal cells to differentiate into osteoblasts, though there is yet no known report of osteoma cutis induced by repeated laser treatment. In summary, we report the first case of Becker's nevus with osteoma cutis, which suggests the possibility of a variable association between Becker's nevus and associated abnormalities. Further studies on the association of Becker's nevus and hamartoma formation will be needed.

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