One person, two bilateral symmetrical giant Becker nevi

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Abstract

Becker nevus, first described by Samuel William Becker in 1949, is a focal epidermal hypermelanotic disorder. It commonly presents as a unilateral hyperpigmented patch that is predominantly distributed on the upper trunk and proximal extremities and frequently associated with hypertrichosis. There have been few reports in the literature of Becker nevus with bilateral involvement; multiple Becker nevi is also unusual. Herein, we report a young man with two bilateral symmetrical giant Becker nevi, one on the trunk with extension to both arms and the second on the abdomen.

Keywords: Becker nevus, multiple, bilateral, symmetrical, giant

Introduction

Becker nevus (BN) is an epidermal melanosis, considered to be an organoid hamartomatous nevus of ectomesodermal tissues [1]. It was first described by Samuel William Becker in 1949, which he described as "concurrent melanosis and hypertrichosis in the distribution of nevus unius lateris" [2]. Characteristically, it is a single unilateral lesion of the upper trunk or proximal extremities, frequently associated with hypertrichosis, and generally manifesting in males during adolescence. However, it may rarely affect other sites, be multiple, or be bilateral [3]. A variety of associated extracutaneous abnormalities have also been described [4,5]. Herein, we present two bilateral symmetrical giant BN in a young man, without any underlying systemic abnormality. In addition, we summarize the previous reports of multiple or bilateral BN, providing a more complete picture of the frequency of these atypical features.

Case Synopsis

A previously healthy 19-year-old man presented to our outpatient department with complaints of large hyperpigmented patches involving the upper anterior chest, back, both arms, and the infra-umbilical region. The small light brown patches were noticed by the parents when he was a 6-year-old. These gradually increased in size, turned darker, and grew excessive hair over the years. There were no associated symptoms or history of a similar problem in the family. Being concerned by its cosmetic appearance, the patient had initiated permanent hair removal a few months earlier.

Cutaneous examination revealed two extensive symmetrical hyperpigmented patches, the largest one involving the upper anterior chest, back, and suprascapular region with extension to both arms, and the other one on the infra-umbilical region measuring about 29cm×15cm in size (**Figure 1**). The patches were irregular bordered and covered by fine hair, except for the chest and back region, which were the areas where permanent hair removal had been performed. A thorough physical examination did not reveal any neurological nor musculoskeletal defect. Routine laboratory investigations, including blood count, and liver and renal function tests were all within the normal range. A cranial magnetic

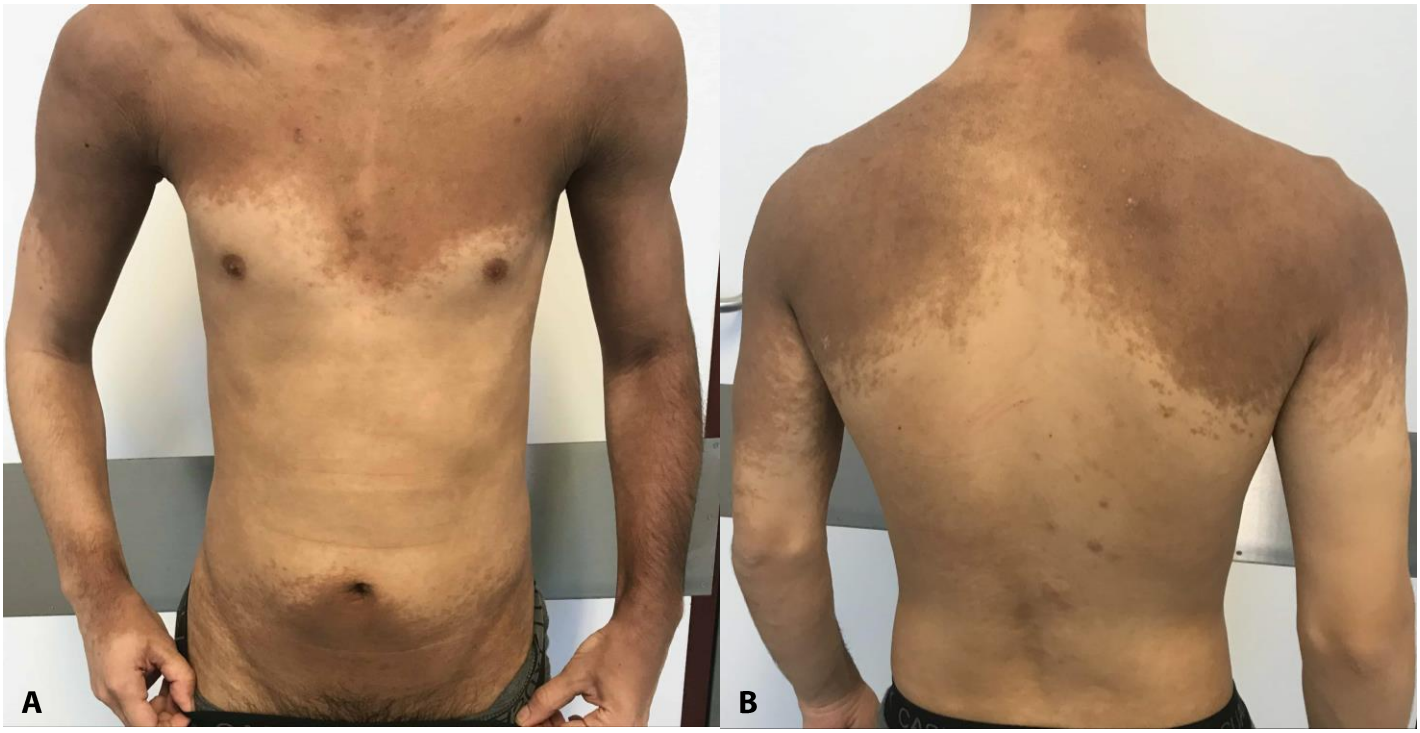

Figure 1. *Becker nevus, clinical picture**: A) and B**) two hyperpigmented patches, one involving the upper anterior chest, back, and suprascapular region with extension to both arms, and the other one located on the infra-umbilical region.*

resonance imaging was also performed, which did not reveal any pathological findings.

The skin biopsy showed a hyperkeratotic epidermis with mild acanthosis, elongated rete ridges, increased pigmentation of basal keratinocytes, and an increase in smooth muscle fibers in the dermis not related to hair follicles. (**Figure 2**).

Both clinical and histopathological findings were consistent with the diagnosis of BN. The patient was diagnosed with two bilateral symmetrical giant BN.

Case Discussion

Becker nevus is a variety of epidermal nevus with dominant clinical features of pigmentation and hypertrichosis. The general prevalence of BN is around 0.5% [6], and it has been described in all races and phototypes. It is about five times more frequent in males, but some authors believe the sex ratio probably does not deviate from unity with the possibility of underreporting in females [1].

Although the etiopathogenesis of BN remains unclear, it is considered to be a hormone-dependent disorder based on the evidence of increased androgen-receptor density and activity in the affected areas [7]. The genetic basis of BN has not been established, but it is assumed to be related to a postzygotic autosomal lethal mutation that survives in a mosaic form [8]. The majority of published cases

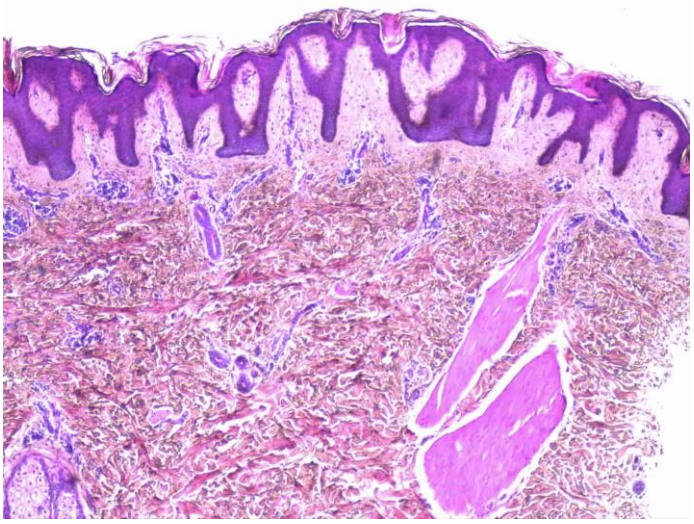

Figure 2. *Becker nevus, histopathological image: microphotograph showing a regular acanthotic epidermis, increased pigmentation of basal keratinocytes and bundles of smooth muscle fibers in the dermis. H&E, 40×.*

of BN are sporadic. However, familial grouping can be observed very rarely, probably in a paradominant inheritance phenomenon [8].

Classically, BN presents as a single unilateral well-demarcated, yet irregularly bordered, tan-to-brown patch, which gradually increases in size, often developing into a geographic pattern [9]. Over time, hypertrichosis typically develops within the lesion. The most common sites involved are the upper anterior chest, back or shoulder, but they can be found on any area of the body, including the forehead, face, neck, lower trunk, extremities, and buttocks [9]. Becker nevus usually presents during puberty, but rare cases of late-onset and congenital BN have been observed [3]. The natural course is for the lesions to persist indefinitely. Various associated extracutaneous abnormalities have been described along with BN, such as breast hypoplasia, aplasia of the pectoralis muscle, shortening limbs, scoliosis, spina bifida, hallux valgus, pectus carinatum, congenital adrenal hyperplasia, and an accessory scrotum [5]. The term Becker nevus syndrome was suggested in 1997 by Happle and Koopman [4], to define the presence of BN in association with ipsilateral breast hypoplasia and ipsilateral musculoskeletal defects. In our patient, no such abnormalities were found, thus ruling out Becker nevus syndrome.

Since the original description of the entity, various atypical clinical presentations of BN have been observed, including atypical sites, uncommon age of onset, and absence of hypertrichosis [3]. However, the findings of multiple BN in a single patient and bilateral involvement have rarely been reported. A careful literature review revealed 26 patients with either one, or both of these two atypical features, summarized in **Table 1**.

There are 16 patients published with multiple BN [5,10-23], the majority of them with bilateral asymmetrical distribution (11 cases), [10-19]. Khaitan et al. [16] described 7 distinct lesions in a 28-year-old man, the highest number reported so far. Li et al. [18] reported a similar case of an extensive area of involvement in a 14-year-old boy with lesions distributed over the trunk and lower limbs, comprising approximately 25% of the total body surface. Remarkably, only two patients have been reported with multiple unilateral BN [22,23] and two patients with multiple bilateral symmetrical BN [5,20]. Hence, to the best of our knowledge, this is the third report of simultaneously multiple and bilateral symmetrical BN. Moreover, four cases have been reported with multiple bilateral asymmetrical BN distributed in checkerboard configuration [16-19], which is characterized by alternating squares with a sharp midline separation. This archetypical pattern represents the type two clinical pattern of cutaneous mosaicism, described by Happle [33], supporting the cutaneous mosaicism theory in the origin of BN.

By contrast, regarding the 10 single bilateral BN reported in the literature [24-32], most of them have symmetrical distribution (8 cases), [24-31] and are usually described as giant roughly symmetric lesions merged in the midline.

Finally, of the 26 patients presented in **Table 1**, 6 had extracutaneous abnormalities, including breast hypoplasia [19, 21], musculoskeletal defects [5,11,19,20,30], mental retardation [5] and cardiac defects [5]. These features do not appear to be correlated with the multiplicity, laterality, symmetry, nor pattern of the BN.

The cosmetic appearance is often the most disturbing feature of BN, as exemplified by our patient. In most cases, lesions are too large to be surgically excised. Potential therapeutic options include electrolysis, waxing, makeup, or laser treatment [6]. Laser therapies in vogue include erbium-yttrium-garnet (Er YAG) laser, and Q-switched laser. Patients with BN with no underlying muscular or skeletal abnormalities should be reassured regarding the benign nature of the condition.

Conclusion

The reported case of two distinct bilateral and symmetrical lesions in a young adult, comprising a large body area of involvement, emphasizes that the spectrum of BN includes lesions without the conventionally associated manifestations of a solitary unilateral hyperpigmented irregular patch.

Contrary to the widely held belief, we wonder whether cases of BN with atypical clinical features are uncommon or underreported.

Potential conflicts of interest

The authors declare no conflicts of interest.

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Table 1. *Published cases of multiple and/or bilateral Becker nevus.*

References	Age (years)	Sex	Localization	Multiplicity	Laterality	Symmetry/ Pattern	Extracutaneous abnormalities
Copeman et al [10]	20	M	Back	Multiple	Bilateral	Asymmetrical	No
	18	M	Back, right arm, left thigh	Multiple	Bilateral	Asymmetrical	No
Thappa et al [11]	34	F	Upper arms, left thigh and chest	Multiple	Bilateral	Asymmetrical	Yes
Bansal et al [12]	18	M	Back	Multiple	Bilateral	Asymmetrical	No
Barman et al [13]	19	F	Right arm and mid back, left knee	Multiple	Bilateral	Asymmetrical	No
Özuğuz et al [14]	15	M	Right lower back, left shoulder	Multiple	Bilateral	Asymmetrical	No
Alhuqayl et al [15]	20	F	Upper back, chest and breasts	Multiple	Bilateral	Asymmetrical	No
Khaitan et al [16]	28	M	Chest, abdomen, back, left arm and forearm, right groin and thigh, both knees and legs	Multiple	Bilateral	Asymmetrical/ checkerboard pattern*	No
Ramot et al [17]	39	M	Left upper back and arm, right chest and lower back	Multiple	Bilateral	Asymmetrical/ checkerboard pattern	No
Li et al [18]	14	M	Trunk, inguinal regions, lower limbs	Multiple	Bilateral	Asymmetrical/ checkerboard pattern	No
Wongtada et al [19]	20	F	Right hemibody, left upper back	Multiple	Bilateral	Asymmetrical/ checkerboard pattern	Yes
Dasegowda et al [5]	15	F	Genitals, thighs, neck	Multiple	Bilateral	Symmetrical*	Yes
Chiramel et al [20]	13	M	Forearms	Multiple	Bilateral	Symmetrical	Yes
De la Torre et al [21]	19	M	Anterior trunk, bilateral arms	Multiple	Bilateral	NM	Yes
Pahwa et al [22]	16	M	Left face and neck	Multiple	Unilateral	-	No
Mehta et al [23]	35	M	Right shoulder and chest	Multiple	Unilateral	-	No
Ferreira et al [24]	4	F	Scapular regions, shoulders, arms	Single	Bilateral	Symmetrical	No
Khatami et al [25]	14	M	Anterior chest, upper abdomen, back, arms and forearms	Single	Bilateral	Symmetrical	No
Grim et al [26]	45	M	Anterior chest	Single	Bilateral	Symmetrical	No
Issa et al [27]	70	M	Trunk, arms	Single	Bilateral	Symmetrical*	No
Yeşilova et al [28]	16	F	Abdomen	Single	Bilateral	Symmetrical	No
Rao AG [29]	23	M	Both shoulders and arms	Single	Bilateral	Symmetrical	No
Bhushan et al [30]	22	M	Both scapulae and shoulder and both upper extremities	Single	Bilateral	Symmetrical*	Yes
Sancheti et al [31]	24	M	Anterior chest, scapular region and both arms	Single	Bilateral	Symmetrical	No
AlGhamdi et al [32]	NM	M	Trunk and proximal right thigh	Single	Bilateral	NM	No
	NM	M	Upper back extending to both arms	Single	Bilateral	NM	No

*Key: No., number; M, male; F, female; NM, not mentioned; *, not mentioned but suggested by text description and/or figures; -, not applicable.*