

Concomitant Kissing Nevus of Penis and Giant Becker's Melanosis of Lower Limb

Eşzamanlı Görülen Penisin Öpüşen Nevüsü ve Alt Ekstremitenin Dev Becker Melanozisi

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ABSTRACT Kissing nevi or split nevi are congenital melanocytic nevi on adjoining areas of the body formed as a result of separation during embryogenesis. Most kissing nevi are seen over the eyelids and only a handful of them have been reported over the penis. Becker's melanosis is a common androgen dependent hamartoma with epidermal, melanocytic or smooth muscle origin. Herein, we describe the case of a 20-year-old male who presented with kissing nevi over the glans penis and prepuce. The patient also had a large Becker's melanosis over the left side extending from the lower back to left lumbar area, the lateral aspect of left buttock and thigh till the knee. Our case is the first instance where a kissing melanocytic nevus is associated with Becker's melanosis.

Keywords: Pigmented nevus; nevus; Becker's nevus

ÖZET Öpüşen nevüsler veya bölünmüş nevüsler vücudun bitişik bölgelerinde embriyogenez sırasında ayrılmanın sonucu gelişen doğumsal melanositik nevüslerdir. Öpüşen nevüslerin çoğu göz kapaklarında görülür ve sadece çok azı peniste bildirilmiştir. Becker melanozisi epidermal, melanositik veya düz kas kaynaklı, yaygın görülen androjen bağımlı bir hamartomdur. Burada glans penis ve prepsiyum üzerinde öpüşen nevüs nedeniyle başvuran 20 yaşında erkek hastayı sunuyoruz. Hastada ayrıca bel bölgesinden başlayıp sol lumbar bölgeye uzanan, sol kalçanın lateralinden dize kadar uzanım gösteren büyük bir Becker melanozisi mevcuttu. Olgumuz öpüşen bir melanositik nevüsün Becker melanozisi ile birliktelik gösterdiği ilk olgudur.

Anahtar Kelimeler: Pigmente nevüs; nevüs; Becker nevüsü

Kissing or divided nevi are congenital melanocytic nevi occurring on adjoining body areas, produced as a result of separation during embryogenesis.¹ They are rare and have been mostly seen over the eyelids. There are only a few case reports of the kissing nevi of the penis. Becker's nevus is a common hyperpigmented hamartoma with features of melanocytic, epidermal and smooth muscle hyperplasia, that usually occurs in adolescence on the upper trunk.² We describe a case of kissing nevus of the penis in a young male with a giant Becker's nevus of the leg, an association never reported before in literature.

CASE REPORT

A 20-year-old male patient presented to us with complaints of gradually progressive darkening of the skin on the left side of the lower trunk and leg since the past 2 years. The lesion was asymptomatic and associated with overgrowth of the hair at the affected site. There was no family history of similar complaints. On examination, a large hyperpigmented plaque with feathery margins was found unilaterally on the left side, extending from the back on the lumbar area including the lateral aspect of the gluteal area and down to the thigh and the knee. The affected area showed

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FIGURE 1: Hyperpigmented plaque with hypertrichosis on a) the left side of lower abdomen and back, lateral aspect of left buttock and thigh and b) left thigh till the left knee.



FIGURE 2: Two hyperpigmented circular patches over the glans penis and inner prepuce.

excessive growth of terminal hair (Figure 1a, Figure 1b). Genital examination revealed two well defined circular hyperpigmented patches, one of size 2 cmx2 cm over the glans extending up to the corona glandis, while the other 1.5 cmx1.5 cm over the lateral aspect of the inner side of prepuce, with the intervening coronal sulcus being completely spared (Figure 2). The two lesions appeared to meet when the prepuce was drawn over the glans. The patient informed that the penile lesions were present for as long as he could remember. Rest of the dermatological and systemic examination was normal.

Routine laboratory investigations including blood counts were normal. Radiograph of the back and thigh

was normal. Skin biopsy from the thigh showed acanthosis, elongation of rete ridges, increased pigmentation of the basal layer and dermal melanophages (Figure 3). The patient did not consent for biopsy from the penile lesions. Based on clinical and histopathological findings, a diagnosis of Becker’s nevus with kissing melanocytic nevus of penis was made.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

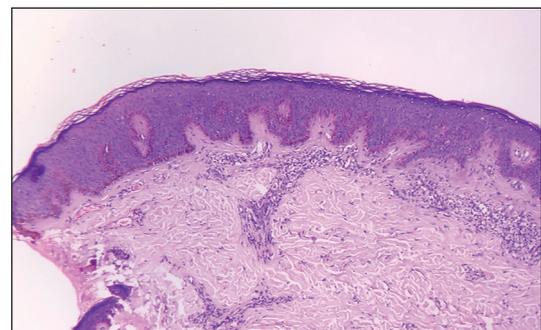


FIGURE 3: Histopathology from the skin of thigh showing acanthosis, elongation of rete ridges, increased pigmentation of basal layer and dermal melanophages. (H&E, 100X).

DISCUSSION

Kissing nevus was first reported on the eyelids and around 40 such cases have been described. Other varieties of kissing nevi include the epidermal nevi, mast cell nevi and nevus spilus.³ Kissing nevi of the penis is very rare; only about 23 cases have been reported so far in the literature.⁴ These lesions show mirror image symmetry and all have been described on the dorsolateral aspect of the glans and the inner surface of the prepuce, with sparing of the coronal sulcus. Divided nevi are believed to be formed prior to the embryonic separation of the adjoining areas during the process of embryogenesis. In the case of penis, the lesion is believed to occur after the migration of the melanoblasts at the 12th week of gestation and before the epithelial placode invaginates and severs to form the prepuce and the glans penis by the end of 13th week of gestation.⁵ The predilection for the dorsal side of the penis is explained by the fact that the epithelial invagination occurs first at the dorsal penis and moves to the ventral aspect.³ Histopathology has shown that these nevi are either intradermal or compound melanocytic nevi. Egberts et al. have reported a case of malignant transformation of kissing penile nevus and therefore, follow up of these patients may be needed.⁶ Aesthetic reasons may warrant treatment, which includes options like surgical excision and skin grafting or laser treatment.¹

Becker's nevus is a common cutaneous hamartoma that is usually seen in young adolescent males and presents as a unilateral hyperpigmented patch on the upper trunk and proximal upper extremities. Becker's nevus in our patient was atypical with respect to its location and unusually large size. Males are affected in a ratio of 5:1 compared to females.⁷ Hypertrichosis is seen in around 50% cases.⁷ It is androgen dependent, suggested by its onset at puberty and presence of hypertrichosis and acneiform lesions at the site. Several musculoskeletal anomalies may be associated with Becker nevus, referred to as Becker nevus syndrome. Becker's nevus has also been associated with several conditions such as pityriasis versicolor, osteoma, lichen planus, nevus depigmentosus, hypohidrosis, prurigo nodularis, granuloma annulare, skin malignancies etc.⁸

Though considered as a type of organoid epidermal nevus by most experts, classification of Becker nevus is controversial. It has been considered by various authors as a type of organoid epidermal nevus, melanocytic nevus or a smooth muscle hamartoma.^{2,9} Histology of Becker's nevus does not show the presence of nevus cells, rather smooth muscle hamartomatous picture may be seen. Eczematous response was described in Becker's nevus in a case report.¹⁰ Eczematous response is otherwise a documented feature of melanocytic nevi, but the cause of eczema in this report was ascribed to the Wolf's isotopic response.

"Twin spotting" or didymosis refers to the occurrence of two different types of nevi and is believed to occur due to postzygotic crossover producing two homozygous daughter cells which represent stem cells of the two distinct types of nevi and other extracutaneous defects. The theory of non-allelic didymosis is no longer accepted as the origin of the paired nevi from a single postzygotic mutation in a heterozygous state was demonstrated in phakomatosis pigmentokeratolica. However, the concept of allelic didymosis proposed for the occurrence of nevus anemicus and nevus flammeus and cutis tricolor may still hold true.¹¹ Rodríguez-Díaz et al. described the occurrence of epidermal nevus and Becker's melanosis in a 16-year-old female and speculated if the co-existence was due to didymosis (which should now be considered as pseudodidymosis).¹²

An association of melanocytic nevus and Becker's melanosis has not been described in literature to the best of our knowledge. More studies are needed to establish if this association was due to pseudodidymosis or chance alone.

Source of Finance

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Mohammad Adil, Syed Suhail Amin; **Design:** Mohammad Adil, Mohd Mohtashim, Roopal Bansal; **Control/Supervision:** Mohammad Adil, Syed Suhail Amin; **Data Collection and/or Processing:** Suhailur Rehman, Prateek Pathak; **Analysis and/or Interpretation:** Mohammad Adil, Suhailur Rehman, Mohd

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