https://doi.org/10.5021/ad.2018.30.2.234

Atypical Proliferative Nodule with Melanocytic Intraepidermal Pagetoid Spreading Arising within a Congenital Melanocytic Nevus in a Pregnant Woman

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Dear Editor:

A 28-year-old woman at the 38th week of her first gestation presented with rapid enlarging papules and nodules in a 15 cm-sized black-pigmented plaque on her right posterior thigh. The plaque had been present since birth, and remained stable in size and pigmentation throughout her life. However, multiple clustered papules and nodules abruptly developed during the 2nd and 3rd trimester of her pregnancy (Fig. 1). To exclude the malignant melanoma arising within a congenital melanocytic nevus (CMN), a punch biopsy was performed. The histopathologic finding showed epidermal proliferation of melanocytes in the background of nevoid cells. The Ki-67 labeling index of the epidermal melanocytes mildly increased as 10% to 20%, whereas dermal melanocytes showed less than 5%. To exclude malignant melanoma, the patient underwent excision of the entire nevus. The distribution of pagetoid cells was mostly confined to the lower part of epidermis, without mitosis, necrosis and high-grade atypia (Fig. 2). The patient finally received the definite diagnosis of benign proliferative nodules (PNs) with mild atypia arising in a CMN.

PNs usually represent the benign nodular proliferation of intradermal melanocytes[1]. PNs arising in a CMN occasionally need to be distinguished from melanoma, because of clinical characteristics such as rapid proliferation, hemorrhage, and ulceration. However, PNs have distinct histopathologic features: non-expanding, blending with the surrounding nevus, maturation, and benign prognosis[2].

Interestingly, our patient presented abruptly, rapidly enlarging PNs during her pregnancy. According to previous studies, the leading opinion is that pregnancy cannot induce significant clinical and dermoscopical changes in melanocytic nevi, except for women with dysplastic nevus syndrome[3]. However, Chan et al.[4] reported that histopathologic changes in melanocytic nevi during pregnancy had tendencies of higher mitotic rates, cellular atypia, and increased proliferation. Although these studies have focused on common nevi or dysplastic nevi rather than CMN, the results could partly explain the abrupt proliferation of atypical nevoid cells during pregnancy in our case.

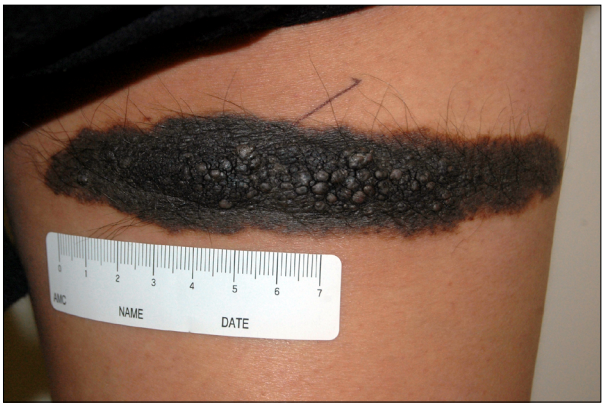

Fig. 1. Multiple papules and nodules arising in a black-colored, slightly elevated plaque on the right posterior thigh during pregnancy.

Received May 27, 2016, Revised March 22, 2017, Accepted for publication March 30, 2017

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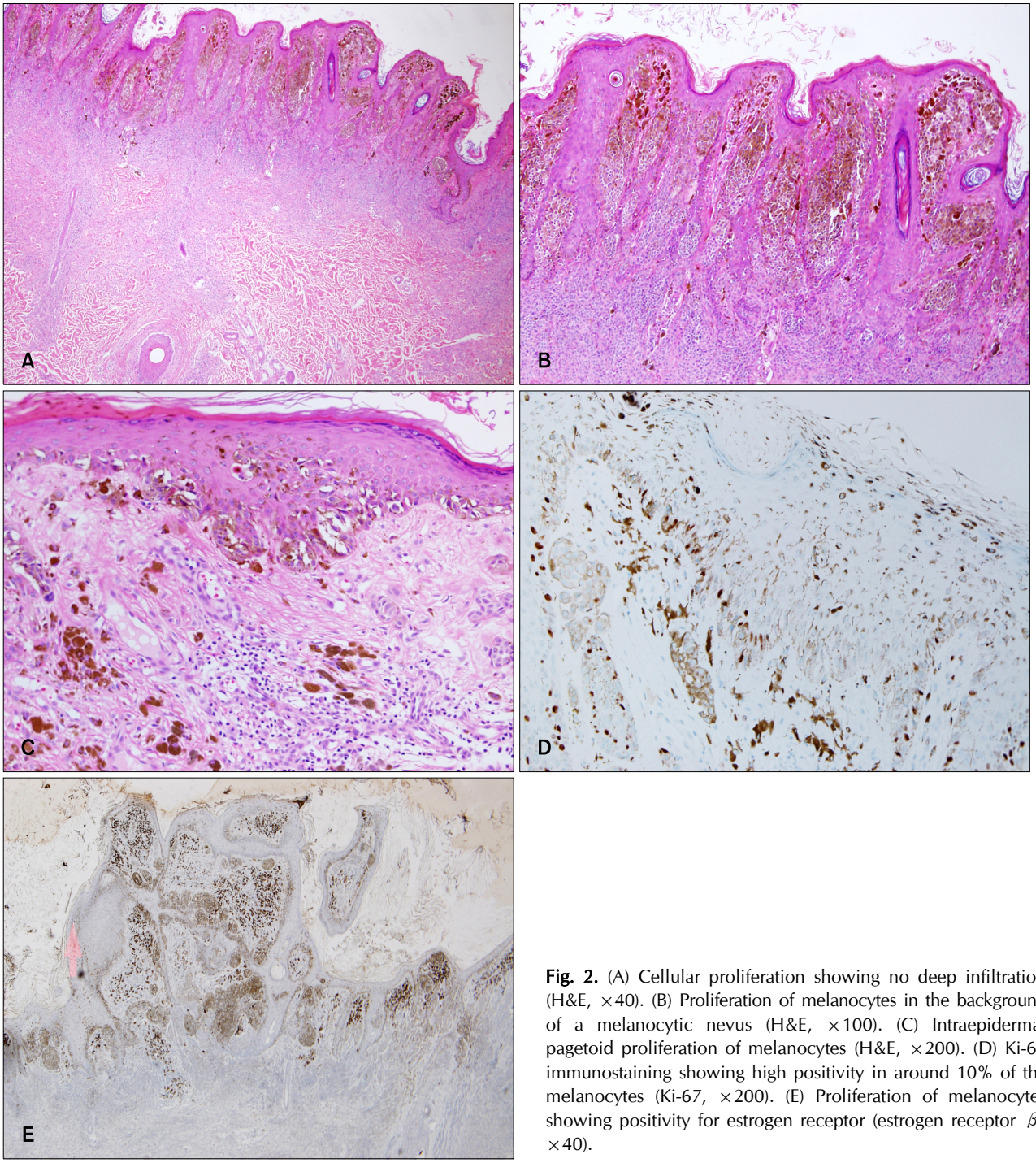

Fig. 2. (A) Cellular proliferation showing no deep infiltration (H&E, ×40). (B) Proliferation of melanocytes in the background of a melanocytic nevus (H&E, ×100). (C) Intraepidermal pagetoid proliferation of melanocytes (H&E, ×200). (D) Ki-67 immunostaining showing high positivity in around 10% of the melanocytes (Ki-67, ×200). (E) Proliferation of melanocytes showing positivity for estrogen receptor (estrogen receptor β, ×40).

The development of PNs in our patient might have been associated with hormonal changes during pregnancy. Estrogen receptor β (ER-β) expression is well known to be found in nevi and malignant melanomas, and to be influenced by hormonal changes in the pregnancy and post-partum periods[3]. Because ER-β suppresses the proliferation of nevoid cells and plays a protective role by inhibiting melanoma transformation, a decreased level of ER-β expression in CMN during pregnancy could be assumed to induce the development of PNs in CMN. However, There is not enough evidence of an association between ER-β expression level and histological atypia during pregnancy[5].

In our case, although aggressive histological features including pagetoid spread of melanocytes and cellular atypia were mimicking malignant melanoma, the final diag-

nosis was benign PNs based on the overall benign architectural features. As shown by this case, the hormonal changes during pregnancy can be associated with various clinical and histopathological changes of CMN. However, even during pregnancy, whenever exclusion of malignant melanoma is required, proper procedures should be performed immediately.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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https://doi.org/10.5021/ad.2018.30.2.236

Scrotal Calcinosis in Brothers

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Dear Editor:

Scrotal calcinosis (SC) is a rare pathological condition characterized by painless, hard, asymptomatic nodules on scrotal skin without any tissue injury or metabolic derangement. The nodules are generally skin-colored, yellowish or white, and consist of calcium and phosphate deposits. SC usually affects patients in childhood or early adulthood. Because of the age of onset and location of occurrence, the condition may be a cause of embarrassment or misunderstanding. We describe the cases of two young adult patients with SC, who were brothers.

Two healthy Korean men, aged 22 and 23 years old, respectively, presented with scrotal nodules that gradually increased over time. The nodules first appeared in adolescence and increased in size and number during the previous 5 to 6 years. Both of the patients denied trauma, associated symptoms, or any prior treatments. There was no history of other systemic inflammatory or metabolic disease. Physical examination revealed multiple firm, non-tender, whitish papules on the scrotums of both patients (Fig. 1A, B). Skin biopsy was performed on both of the patients, and multiple calcium deposits and basophilic globules were found in the dermis (Fig. 1C, D). Routine laboratory examinations including serum calcium and phosphorus were all within normal limits. The possibility of tumoral calcinosis was ruled out, as both the patients had rela-

Received January 9, 2017, Revised March 7, 2017, Accepted for publication April 3, 2017

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