Two Cases of Pigmented Bowen's Disease

Seon-Wook Hwang, M.D., Jung-Wook Kim, M.D., Sung-Wook Park, M.D., Han-Young Wang, M.D.

Department of Dermatology, College of Medicine, Inje University, Busan, Korea

Pigmented Bowen's disease(PBD) is a rare variant of Bowen's disease(BD). Most of the reported cases showed pigmented patches or thin plaques. Thus its clinical manifestations may simulate other various pigmented skin lesions. We experienced 2 cases of PBD in patients with multiple BD developed after taking Korean proprietary pills(KPP, "Hwan-Yak"), which were suspected to contain certain amount of arsenics. Both patients also showed arsenical keratosis on their palms and soles. The darker pigmentation of the PBD led us to differentiate them from melanoma. (Ann Dermatol 14(2) 127-129, 2002).

Key Words : Pigmented Bowen's disease

Pigmented Bowen's disease(PBD) is almost identical to clinical and histopathologic findings of Bowen's disease(BD) except for various degree of melanin deposition[1]. After Lloyd[2] had reported a case of so-called multicentric pigmented Bowen's disease, many of the pigmented lesions reported in the anogenital area as Bowen's disease would now be regarded as examples of bowenoid papulosis[3]. The only pigmented skin lesions outside of the anogenital area that show histopathologic changes similar to that of Bowen's disease are now generally accepted as PBD. But the reports of such a case were not common even worldwide. Ragi et al[3] reviewed the records of 420 cases of BD and found seven(1.7%) to be pigmented, none of which involved the anogenital area. In our review, only one case of PBD have been reported in Korean dermatological literature[4]. The clinical presentation of the previously reported PBD cases were similar to the classic BD except for variable degrees of pigmentation[3]. However, even PBD rarely shows severe pigmentation. In that case it should be differentiated from melanoma.

We experienced 2 cases of PBD in patients with past history of long-term arsenic pill therapy. They showed dark pigmented plaques that were very similar to melanoma. These findings seem to be uncommon and interesting.

Received August 1, 2001.
Accepted for publication October 20, 2001.
Reprint request to : Seon-Wook Hwang, M.D., Department of Dermatology, Pusan Paik Foundation Hospital, 633-165, Kaekeum-Dong, Pusanjin-Ku, Pusan 614-735, Korea
Tel. (051)890-6391, Fax. (051)893-7233
E-mail : mompoo@lycos.co.kr

CASE REPORTS

Case 1

A 72-year-old man presented with a black plaque on the dorsum of his right foot for 1 year (Fig 1). He also presented with skin colored keratotic papules on his palms and soles. He took Korean proprietary pills(KPP, "Hwan-Yak") which were suspected to contain certain amount of arsenics for various medical problems, but he could not recall the exact amount and duration of the intake. About ten years ago, the hyperkeratotic papules developed on his palms and soles. On physical examinations, the black plaque lesion measured about 1.5cm × 1.5cm. The whole lesion was excised completely. Histopathological examination revealed epidermal changes of BD such as hyperkeratosis, parakeratosis and dyskeratosis. Severe acanthosis was also observed. Apparently atypical keratinocytes were arranged haphazardly and contained opulent pigments (Fig 2). There was no evidence of dermal invasion. Upper dermis contained

Annals of Dermatology
Vol. 14, No. 2, April 2002

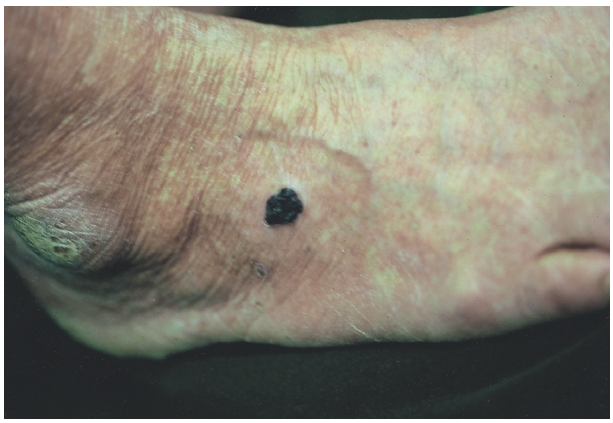

Fig. 1. Case 1. About 1.5×1.5cm sized black nodule on the dorsum of right foot.

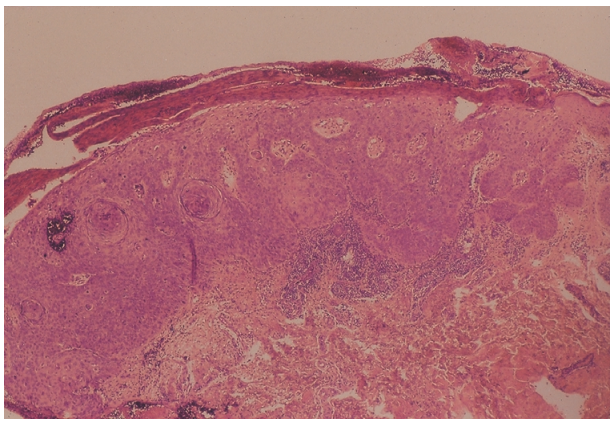

Fig. 2. Case 1. Atypical keratinocyte were arranged haphazardly and contained pigments.

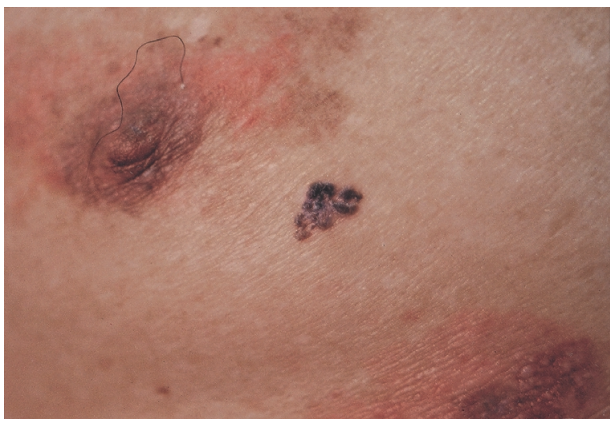

Fig. 3. Case 2. Grouped black papules on right chest.

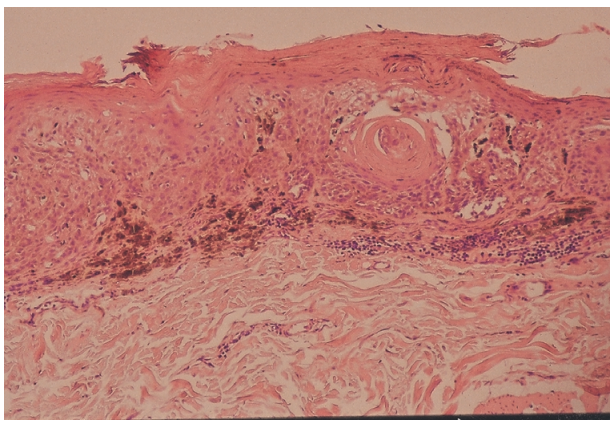

Fig. 4. Case 2. Pigment-containing atypical keratinocytes in all layers of the epidermis (H&E ×100).

many lymphocytes and melanophages.

Case 2

A 55-year-old man visited our department for the evaluation of scaly erythematous patches and diffuse macular pigmentations on the whole body. He also presented with grouped black papules on his right chest(Fig 3) and hyperkeratotic papules on his palms and soles. About 40 years ago, he took Korean proprietary medical pills(KPP, "Hwan-Yak") that was suspected to contain arsenic for his facial laceration wound. Ten years later, the hyperkeratotic papules developed on his palms and soles. About six years ago, a diffuse macular pigmentation developed on his trunk and extremities. And the numbers of these pigmentations increased slowly. About three years ago, the scaly multiple erythematous patches on the extremities, and an indurated black plaque on the right chest appeared. The scaly erythematous pathes were scattered on both thigh and calf in variable sizes. The black plaque lesion comprised several small indurated papules and it measured about 1.0cm×1.5cm. Excisional biopsy was done. Histopathological examination revealed pigment-containing atypical keratinocytes in all layers of the epidermis and many melanophages in the papillary dermis(Fig 4). The other findings were similar to that of the first case. And the scaly erythematous skin lesions on the extremities were revealed as multiple Bowen's disease.

DISCUSSION

The 'pigmented' Bowen's disease, which had

been reported first by Yet et al[5], is ordinary BD lesion with hyperpigmentation. Its clinical features were described as well-demarcated, scaly erythematous patches or thin plaques with pigmentation. The histopathological degree of pigmentation was not mentioned.

Chronic arsenism is a systemic disease resulted from chronic exposure to arsenic in the form of metal itself, inorganic arsenic and organic arsenic[6]. Various manifestations in the human body due to arsenism are described already. In the field of dermatology, skin problems such as arsenical keratosis, multiple Bowen's disease and dyspigmentary changes are taken seriously[7].

Our patients took certain pills that were suspected to contain arsenic when they were young. About thirty to forty years later, black plaque lesions of different size developed in addition to the other various arsenic-related skin lesions including diffuse hyperpigmentation, BD and arsenical keratosis. The clinical features of the black plaques were similar to that of malignant melanoma. The lesions were located on the dorsum of the foot in one patient and on the chest in the other. Histopathological examination showed epidermal hyperkeratosis, parakeratosis, dyskeratosis, acanthosis and atypia of keratinocytes. There were also precipitations of pigments in all layers of epidermis. Many dermal melanophages were present. According to the above findings it may be certain that our cases are caused by chronic exposure to arsenic which is notorious for its potentiality for pigmentary alteration. We also examined the classic multiple BD lesions of the one patient. But these lesions were not pigmented clinically and histopathologically despite the fact that the patient showed hyperpigmentation of the whole body.

The mechanism of dyspigmentation in chronic arsenism has not been sufficiently understood. Some heavy metals cause hyperpigmentation by being deposited difffusely in the dermis, but arsenic is supposed to cause hyperpigmentation by altering melanin deposition. The increased melanin production resulted from its interactions with melanocytes[8]. For interactions of melanocytes, Burgoyne et al[9] had shown that melanocyte hyperplasia and increased melanin synthesis might cause hyperplasia of epidermal cells. Pinkus et al[10] insisted that some symbiotic relation of melanocytes and epidermal cells caused pigmentary variation in a certain pathologic condition. Thus it seems that this characteristic interaction between melanocytes, epidermal cells and carcinogens might play a major role in determining the morphologic patterns of dyspigmentation[3].

Our cases have characteristic clinical features that are different from the ordinary PBD lesions. The differences include their melanoma-like clinical features, the epidermis containing a lot of pigments and co-existence with classic, non-pigmented BD lesions in the same patients. It is supposed that the mechanism of hyperpigmentation in the PBD of our patients cannot be soley illustrated by chronic arsenic ingestion. And these PBD lesions should be taken into account in differential diagnosis of malignant melanoma, seborrheic keratosis and dysplastic nevus.

REFERENCES

1. Weedon D: Tumors of the epidermis. In Weedon D, Strutton G (eds): Skin Pathology. Churchill Livingstone, New York, 1997, pp645-646.
2. Lloyd KM: Multicentric pigmented Bowen's disease of the groin. Arch Dermatol 101:48-51, 1970.
3. Ragi G, Turner MS, Klein LE, Stoll HL Jr.: Pigmented Bowen's disease and review of 420 Bowen's disease lesions. J Dermatol Surg Oncol 14:765-769, 1988.
4. Lee HJ, Oh ST, Ha SJ, Kim JW: A case of pigmented Bowen's disease. Ann Dermatol 12:68-70, 2000.
5. Yeh S, How SW, Lin CS: Arsenical cancer of skin. Cancer 21:312-339, 1968.
6. Chen CJ, Kuo TL, Wu MM. Arsenic and cancer. Lancet 1988;1:414-415
7. Wong SS, Tan KC, Goh CL. Cutaneous manifestations of chronic arsenism: Review of seventeen cases. J Am Acad Dermatol 1988;38:179-185
8. Levantine A, Almeyda J. Drug induced changes in pigmentation. Br J Dermatol 1973;89:105-112
9. Burgoyne FH, Heston WE, Hartwell JL, Stewart HL. Cutaneous melanin production in mice following application of the carcinogen 5,9,10 trimethoyl-1,2 benzanthracene. J Natl Cancer Inst 1949;10:665-668
10. Pinkus H, Staricco RJ, Kropp PJ.: The symbiosis of melanocytes and human epidermis under normal and abnormal condition. In gordon M (ed): Pigment Cell Biology. New York, 1959, pp127.