Case Report of a Late Onset Bowenoid Papulosis Progressing to Squamous Cell Carcinoma: Letter to the Editor

Skuamöz Hücreli Kansere Dönüşen Geç Başlangıçlı Bowenoid Papulozis Olgusu

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Bowenoid papulosis (BP) was first described as a condition affecting the groin by Lloyd, but was recognized as an entity by Kopf and coworkers a few years later. It consists of indolent, usually asymptomatic and pigmented verrucous papules involving the genitalia or perigenital areas of both sexes.[1,2] It is usually caused by human papilloma virus. Often identified are the subtypes 16 and 18.[3] Histological features suggest a preinvasive carcinoma in situ, but the course of the disease is usually benign.[2] BP usually behaves like genital warts on the external genitalia. On the glans penis of an uncircumcised male and on the cervical, vaginal, or rectal mucosa, progression to invasive squamous cell carcinoma may occur. Women with BP and the female partners of men with BP have a risk of cervical dysplasia.[4]

An 81-year-old male patient presented to our outpatient clinic with an itchy wound on the genital region for one month. His dermatological examination revealed a central 4 x 3 cm hypopigmented plaque surrounded with hyperpigmented papules and a 1 x 1 cm ulcerated tumoral lesion on the ventral side of the penis shaft (Figure 1). The patient was consulted with the Department of Plastic and Reconstructive Surgery. A total excision followed by a closure with skin graft was performed. Histopathological examination of the specimen revealed tumoral lesion consisting of pleomorphic cells with wide eosinophilic cytoplasm, round vesiculated nuclei and distinct nucleoli, superficial orthokeratosis, disorganization, atypical and individual cell keratinization, bursting into bud from basal cells to dermis and chronic inflammatory cell infiltration (Figure 2, 3). Based on clinical and histopathological findings, the patient was diagnosed with BP and poorly differentiated squamous cell carcinoma. The patient was not immunosuppressed. Both the physical examination and the laboratory findings were normal. Recurrence was not observed throughout the follow-up period.

BP has also been described as multicentric pigmented Bowen's disease, which may be referred as pigmented penile papules in men and as re-

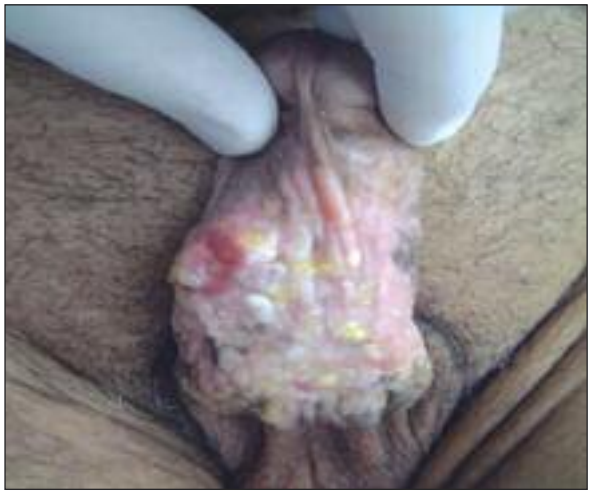

FIGURE 1: Hypopigmented plaque 4 x 3 cm in size at the center with hyperpigmented papules around and an ulcerated tumoral lesion 1 x 1 cm in size on the ventral penis shaft.

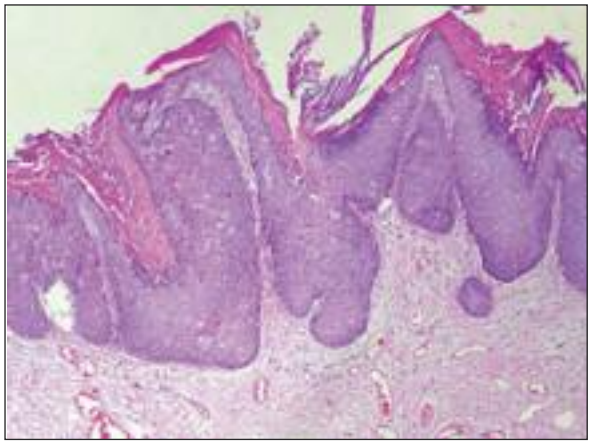

FIGURE 2: Tumoral lesion consisting of pleomorphic cells with wide eosinophilic cytoplasm, round vesiculated nuclei and distinct nucleoli in epidermis (HE, x100).

versible vulvar atypia in women. It characteristically develops in young adults, but has also been described in patients older than 60 years. The initial lesions are multicentric shiny or verrucous papules, which often have an irregular outline with a flat top in some cases. They occur most commonly on the penile shaft or the labia major and may coalesce to form wide velvety plaques over the anogenital skin. Lesions range in colour from reddish-brown to black and may be pruritic.[5] BP has a benign outcome in general and in many instances spontaneous regression has been described for both sexes.[2,5,6] A small number of cases may progress to squamous cell carcinoma (SCC).[3] Human papilloma virus (HPV)-16, HPV-18, HPV-31, HPV-33, HPV-39 and HPV-52 are considered to be highly oncogenic for the development of genital cancers.[2,7] Immunocompromised individuals with BP are at higher risk to develop cancer.[8]

There are a few case reports about BP transforming to squamous cell carcinoma in the literature.[2,8,9] Yoneta et al reported a 35-year-old man with warty papules and plaques on his perianal and genital areas, which had started 8 years ago and had developed superficial ulcers 4 years after the onset of the symptoms. The lesions had recurred after all therapeutic approaches. Both the perianal and left inguinal lesions revealed an invasive squamous cell carcinoma on histology.[2] Hama et al described a 50-year-old woman presenting with asymptomatic, multiple black macular lesions on the genitalia for 6 months. She had multiple, round to irregularly shaped, black macular lesions on her labia major and perineum. The histological diagnoses were BP and SCC. The HPV DNA in SCC and BP was shown to be HPV-31. They concluded that the elevated amount of HPV-31 DNA could have led to the emergence of SCC from BP under the condition of decreased cellular immunity.[9]

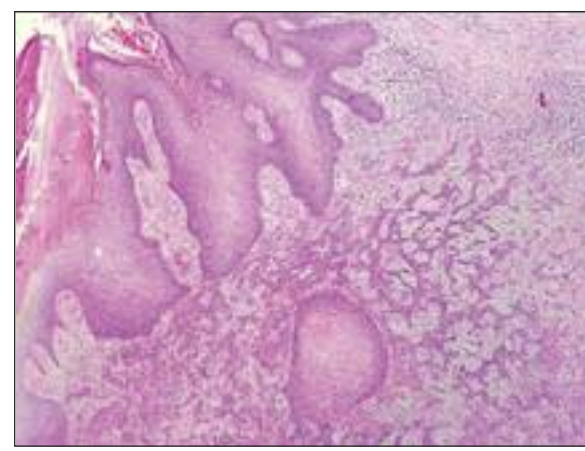

FIGURE 3: Superficial orthokeratosis, disorganization, atypical and individual cell keratinization, bursting into bud from basal cells to dermis with chronic inflammatory cell infiltration (HE, x100).

The presented case was considered interesting as he was very old and was not immunosuppressed. With this interesting case, we wanted to draw attention once more that BP might transform into SCC.

We suggest that the patients diagnosed with BP should be followed-up periodically for the development of SCC even though the lesions were treated destructively. Their sexual partners should also be evaluated for the risk of anogenital neoplasia. We also recommend that HPV vaccine should be routinely used to prevent HPV infections, genital warts, BP and the related malignant conditions.

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