Pigmented Bowen's Disease: Atypical Presentation of a Rare Variant

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Abstract

Bowen's disease is a type of precancerous lesion occurring mainly in older individuals. Multicentric pigmented Bowen's disease is a rare variant with only a few cases reported worldwide. The main objective of this case report is to create an awareness about this unusual variant, its occurrence in a multicentric fashion, and the significance of taking a biopsy to exclude other pigmented lesions. We report the case of a 74-year-old male who presented with multiple warty verrucous lesions on his back, clinically diagnosed as verruca vulgaris. Histopathological examination, which is the gold standard clinched the diagnosis. All pigmented lesions should thus be viewed with suspicion and closely monitored for progression to invasive squamous cell carcinoma. Rapid increases in size with ulceration should raise concern for malignant transformation, and prompt surgical intervention should be considered early to prevent metastases.

Keywords: Precancerous Conditions; Carcinoma, Squamous Cell.

Introduction

Bowen's disease is a rare type of precancerous lesion, occurring mainly on the hair bearing skin in the older individuals. It was first described by Bowen in 1912 as an atypical epithelial proliferative lesion.[1] Review of literature has shown pigmented Bowen's disease to account for less than 2% of all Bowen's disease cases, with very few cases of multicentric pigmented Bowen's reported worldwide.[2] It closely mimics other pigmented benign and malignant lesions like seborrheic keratosis, melanocytic nevus, pigmented basal cell carcinoma, bowenoid papulosis, and malignant melanoma.[3] Histopathologic examination is the gold standard for arriving at a conclusive diagnosis and should be considered in the differential diagnosis of all pigmented lesions. The main objective of this case report is to create an awareness about the unusual pigmented variant of Bowen's disease and the significance of taking a biopsy to exclude other pigmented lesions like basal cell carcinoma and malignant melanoma.

Case Report

A 74-year-old man presented with a two-year history of warty lesions on his back. The lesions, which were asymptomatic, increased in size and number over the years. His medical history was unremarkable. The patient had no history of chronic arsenic exposure or sunburn. On physical examination, there were multiple erythematous papules with central verrucous plaques arranged in annular multicentric fashion on the back [Figure 1]. The clinical differential diagnosis, based on the history and physical examination, included verruca vulgaris and basal cell carcinoma. Histological examination showed full thickness epidermal dysplasia and atypical keratinocytes arranged in a disorderly fashion throughout the epidermis [Figure 2]. Increased melanin pigment was noted in the basal layer of the epidermis [Figure 3 and 4]. The histologic features were consistent with pigmented Bowen's disease. Our patient subsequently underwent

wide local resection of the lesion and no recurrences were documented at the end of eight months follow-up.

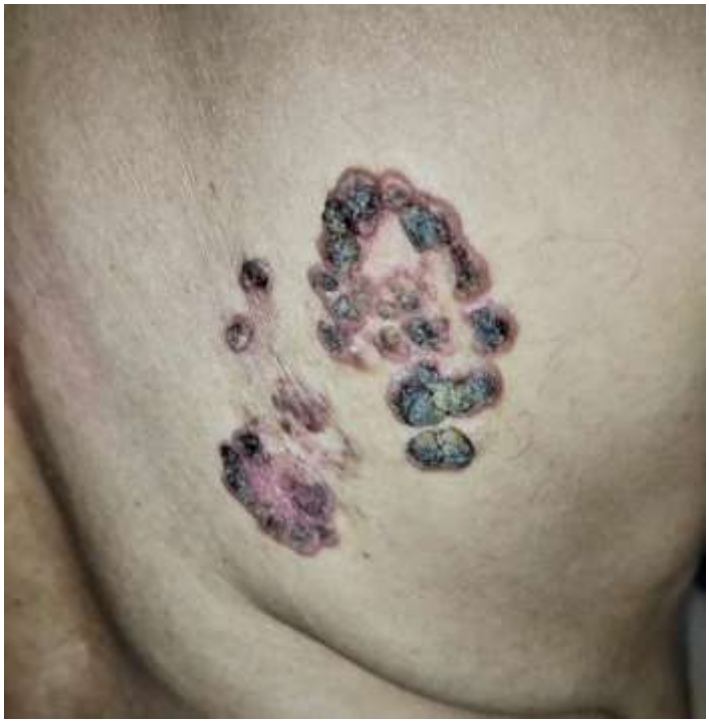

Figure 1: Multiple annular erythematous and verrucous papules and plaques over sun-protected areas on the back.

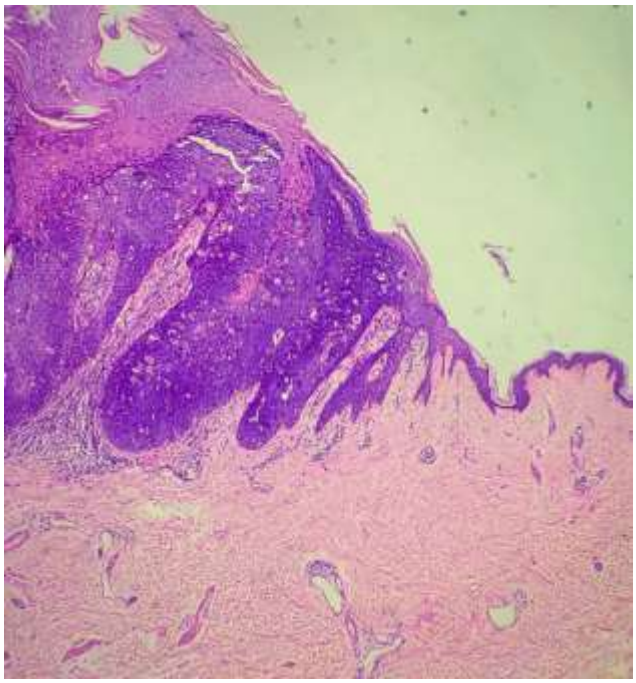

Figure 2: Hematoxylin and eosin staining showing full-thickness keratinocyte atypia with apoptotic cells, dyskeratosis, and an intact basal layer. Magnification = 100 ×.

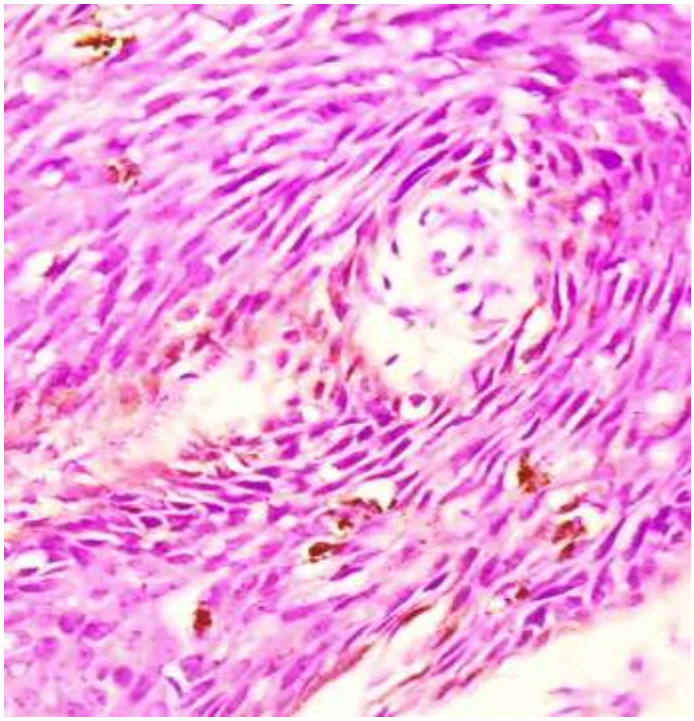

Figure 3: Hematoxylin and eosin staining showing increased melanin pigmentation. Magnification = 400 ×.

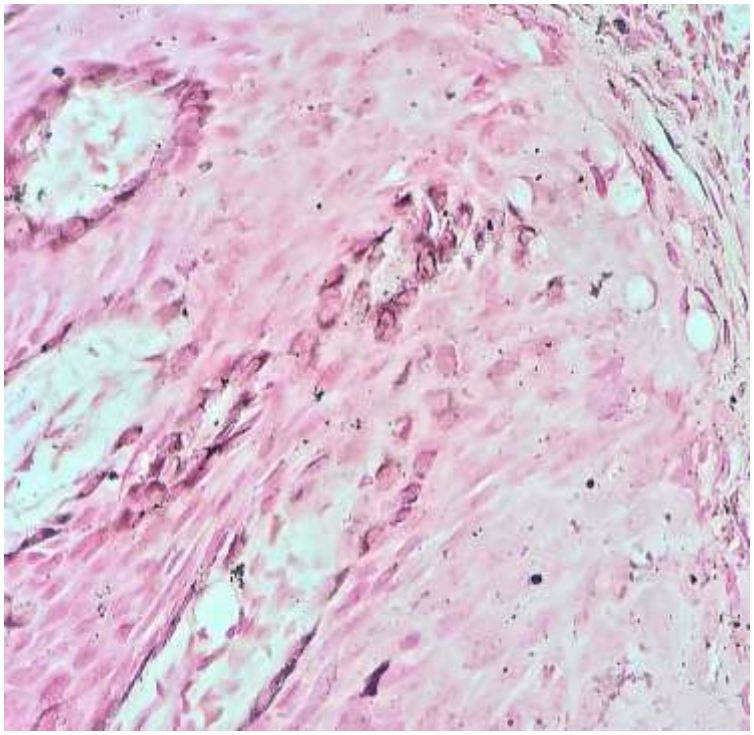

Figure 4: Fontana Masson stain highlighting the melanin pigment. Magnification = 400x

Discussion

Bowen's disease is an unusual type of precancerous lesion with intraepithelial squamous cell carcinoma in situ morphology. It was first described by Bowen in 1912 as an atypical epithelial proliferative lesion.[1] Although Bowen's disease per se has no metastatic potential and exhibits slow growth, approximately 3% cases can progress to full blown invasive squamous cell carcinoma if left untreated.[4] The pigmented variant of Bowen's is even rarer, accounting for less than 2% of all Bowen's cases and its diagnosis is quite challenging as it closely mimics superficial spreading melanoma.[5] In a study that included 420 cases, only 1.67% were pigmented, emphasizing the significance of having a clinical suspicion of pigmented Bowen's in every pigmented lesion.[2] These lesions can at times occur extensively, involving multiple sites or as multiple lesions around a main lesion, as in our case.[6]

The main etiological factors implicated in the pathogenesis of Bowen's disease are sunlight, chronic arsenic exposure, viral infection, trauma, and ionizing radiation. Most cases of Bowen's, particularly those occurring in the anogenital areas, are associated with the oncogenic serotypes of HPV, notably 16, 18, 31, and 33.[7] However, there is also increasing evidence to support the role of HPV in the pathogenesis of extragenital Bowen's disease, particularly in the immunocompromised.[8]

While the lesions in Caucasians are seen to occur in sun-exposed areas, those occurring in black individuals mainly occur in areas unexposed to sun, like the anogenital area, palms, soles, and mucous membranes. The reason for pigmentation in Bowen's has not been fully elucidated; however there are theories supporting the fact that cytokines and inflammatory mediators produced by tumor cells induce melanocytic proliferation and stimulate melanin production. The lesions can range from small discrete brownish eruptions to confluent blackish papules, some of which may be macerated. Some may present as hyperpigmented plaques with a velvety surface. The color of pigmented Bowen's disease varies from brown to black and they may at times be found in association with seborrheic keratosis and solar lentigo. Hyperpigmentation is more commonly observed in lesions occurring in the anogenital area, probably due to the higher temperature found in this region.[9] Bowen's disease is a precancerous lesion most commonly encountered in the older age group, with very few cases occurring below 30 years. There are, however, reports of Bowen's disease in children infected with HIV.[10]

It has a strong predilection for hair-bearing skin, with occasional cases occurring on palms, soles, and mucosa. Bleeker et al., emphasized the significance of carefully monitoring cases of genital Bowen's disease to look for progression to penile cancer.[7] Occasionally, collision tumors may be found, with the simultaneous occurrence of two different tumors at the same anatomical site, where the prognosis is determined by the most malignant tumor. Additionally, this precancerous lesion can at times be masked by other coexistent pigmented lesions or purpura. The lesions are usually asymptomatic, although some may present with pruritus and a burning sensation. The dermoscopic appearance of Bowen's disease can also mimic other pigmented lesions, hence it is no longer considered a reliable investigation to distinguish it from similar other lesions.[9] Hence the gold standard for diagnosing Bowen's disease is by biopsy, which should be done in every chronic pigmented lesion refractory to the usual therapeutic modalities.

Histological features include significant full-thickness dysplasia, loss of polarity, dyskeratotic cells, atypical mitotic figures, and an increase in melanin pigment in the basal layer of the epidermis and upper dermis.[2] The papillary dermis and upper reticular dermis also show marked fibrosis and scattered melanophages. HPV has been incriminated in its pathogenesis, especially the high-risk types.

The various differential diagnosis considered for pigmented Bowen's disease include both benign and malignant pigmented lesions like seborrheic keratosis, melanocytic nevus, pigmented basal cell carcinoma, bowenoid papulosis, and malignant melanoma.[3] It is often a challenging diagnosis to many clinicians who find it extremely difficult to distinguish it from melanoma variants.[11] The diagnosis can therefore only be confirmed by histopathological examination. Consequently, pigmented Bowen's disease is indeed a diagnostic and therapeutic dilemma for most dermatologists. The other available therapeutic options are cryotherapy, topical 5-fluorouracil, electrocautery, and laser therapy.[12] In a study conducted at a referral hospital in Korea, therapeutic efficacy was found to be highest in patients who underwent surgical excision (100%) while recurrence was highest in the 5-fluorouracil group (33.33%).[12]

Conclusion

Bowen's disease should be included as a differential diagnosis for any chronic pigmented lesion of skin. In view of the nonspecific clinical and dermoscopic findings, our case highlights the importance of skin biopsy to confirm the diagnosis and rule out the possibility of an invasive carcinoma. Though Bowen's disease presenting as a solitary lesion is common, occurrence in a multicentric fashion is rare and infrequently reported.

Disclosure

The authors declared no conflicts of interest. Informed consent was obtained from the patient.

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