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

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Received: 21 January 2024

Accepted: 21 April 2024

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DOI 10.5001/omj.2026.34

Abstract

Bowen's disease is a type of precancerous lesion occurring mainly in the older age group. Multicentric pigmented Bowen's disease is a rare variant with only few cases reported worldwide. 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 over back which was 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 to look for progression to invasive squamous cell carcinoma. Rapid increase in size with ulceration should raise concern for malignant transformation and prompt surgical intervention should be considered sufficiently early to prevent metastases.

Keywords: Bowen's; Multicentric; Pigmented.

Introduction

Bowen's disease is a rare type of precancerous lesion occurring mainly on the hair bearing skin in the older age group. It was first described by J.T. Bowen in the year 1912 as an atypical epithelial proliferative lesion.[1] Review of literature has shown pigmented Bowen's to account for less than 2% of all Bowen's disease 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. Therefore, it should be considered in the differential diagnosis of all pigmented lesions. 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 2-year history of warty lesions over the back. The lesions which were asymptomatic, increased in size and number over the years. His past 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 over the back [Figure 1]. The clinical differential diagnosis considered based on the history and physical examination were verruca vulgaris and basal cell carcinoma. Histologic examination showed full thickness epidermal dysplasia and atypical keratinocytes arranged in a disorderly fashion throughout the epidermis. Increased melanin pigment was noted in the basal layer of the epidermis [Figure 2]. The histologic features were consistent with pigmented Bowen's disease. Our patient subsequently underwent wide local resection of the lesion [Figure 3] and no recurrences were documented at the end of 8 months of follow up.

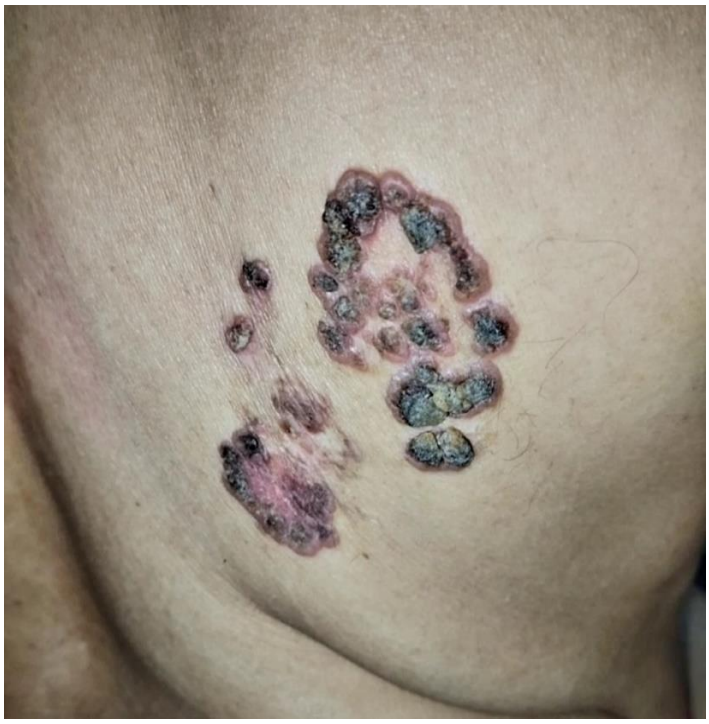

Figure 1: Multiple annular erythematous and verrucous papules and plaques over sun protected areas in the back.

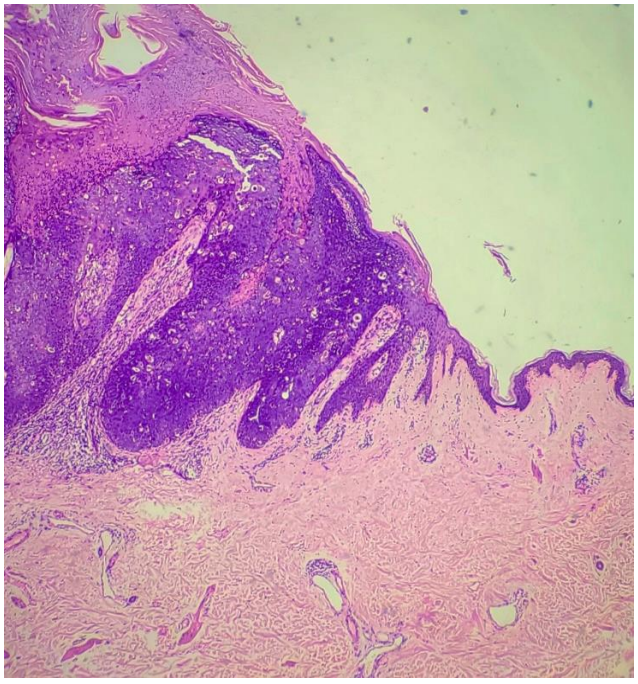

Figure 2: H &E 100 x showing full thickness keratinocyte atypia with apoptotic cells, dyskeratosis and an intact basal layer.

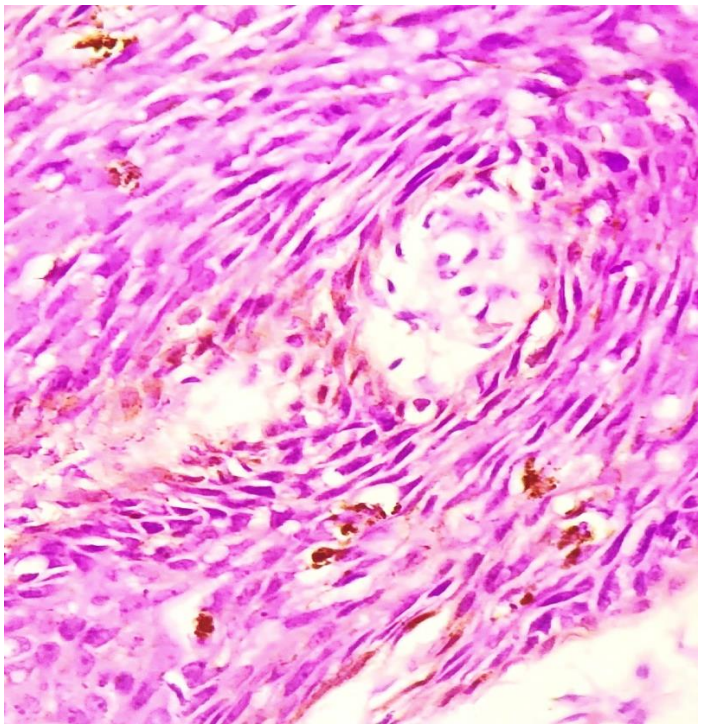

Figure 3: H&E 400x showing increased melanin pigmentation

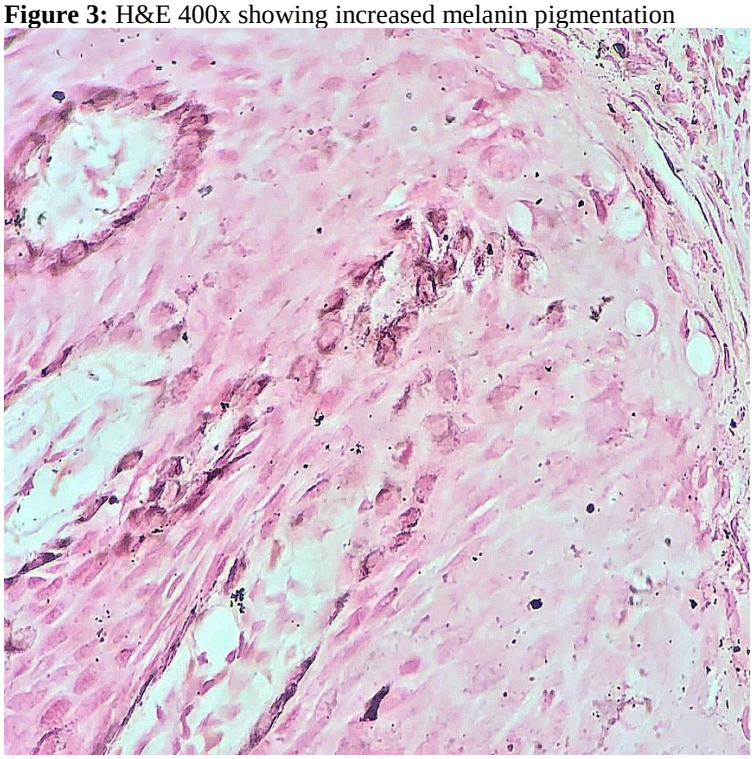

Figure 4: Masson Fontana highlighting the melanin pigment.

Discussion

Bowen's disease is an unusual type of precancerous lesion with intraepithelial squamous cell carcinoma in situ morphology. It was first described by J.T. Bowen in the year 1912 as an atypical epithelial proliferative lesion.[1] Though 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] Pigmented variant of Bowen's is still 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] Ragi et al in their study found that of the 420 cases they examined only 1.67% were pigmented and they emphasized the significance of having a clinical suspicion of pigmented Bowen's in every pigmented lesion.[2] These lesions can at times occur in an extensive manner involving multiple sites or as multiple lesions around a main lesion as in our case.[6]

The main etiological factors incriminated in the pathogenesis of Bowen's 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 the sun exposed areas, those occurring in blacks 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 of them may present as hyperpigmented plaques with a velvety surface. The color of pigmented Bowen's is seen to vary 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 30yrs. There are however reports of Bowen's disease in children infected with human immune deficiency virus (HIV).[10]

It has a strong predeliction for the hair bearing skin with occasional cases occurring on palms, soles and mucosa. Bleeker et al in their study emphasized the significance of carefully monitoring cases of genital Bowen's to look for progression to penile cancer.[7] Occasionally, collision tumors may be found with simultaneous occurrence of two different tumors at the same anatomical site, where the prognosis is determined by the most malignant tumor. Also, this precancerous lesion can at times be masked by other coexistent pigmented lesions or purpura. The lesions are usually asymptomatic, however some may present with pruritus and burning sensation. Dermoscopic appearance of Bowen's disease can also mimic other pigmented lesions, hence 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 therapeautic modalities.

Histological features include significant full thickness dysplasia, loss of polarity, dyskeratotic cells, atypical mitotic figures and increase in melanin pigment in the basal layer of 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 in a pigmented variety of Bowen's 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 nightmare and challenging diagnosis to many clinicians who find it extremely difficult to distinguish it from melanoma variants.[11] The diagnosis can hence be confirmed by histopathological examination only. Therefore, pigmented Bowen's disease is indeed a diagnostic and therapeautic dilemma to most dermatologists. The other available therapeautic 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]

References

1. Bowen JT. Centennial paper. May 1912 (J Cutan Dis Syph 1912;30:241-255). Precancerous dermatoses: a study of two cases of chronic atypical epithelial proliferation. By John T. Bowen, M.D., Boston. Arch Dermatol 1983 Mar;119(3):243-260. Accessed 29 Apr 2024. Internet.

2. 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 1988 Jul;14(7):765-769.

3. Krishnan R, Lewis A, Orengo IF, Rosen T. Pigmented Bowen's disease (squamous cell carcinoma in situ): a mimic of malignant melanoma. Dermatol Surg 2001 Jul;27(7):673-674. Accessed 29 Apr 2024. Internet.

4. Bath-Hextall FJ, Askew DA, Wilkinson D, Leonardi-Bee J. Interventions for Bowen's Disease. Queensland's institutional digital repository (The University of Queensland). 2008 Jul 16

5. Stante M, de Giorgi V, Massi D, Chiarugi A, Carli P. Pigmented Bowen's disease mimicking cutaneous melanoma: clinical and dermoscopic aspects. Dermatol Surg 2004 Apr;30(4 Pt 1):541-544.

6. Kimura S, Hirai A, Harada R, Nagashima M. So-called multicentric pigmented Bowen's disease. Report of a case and a possible etiologic role of human papilloma virus. Dermatologica 1978;157(4):229-237.

7. Zhang L, Han W, Shen G. Bowen's disease with multiple lesions of the penis and scrotum: A case report. Chinese journal of plastic and reconstructive surgery. 2022 Jun 1;4(2):79–81.

8. Švajdler M Jr, Mezencev R, Kašpírková J, Kacerovská D, Kazakov DV, Ondič O, et al. Human Papillomavirus Infection and p16 Expression in Extragenital/Extraungual Bowen Disease in Immunocompromised Patients. Am J Dermatopathol 2016 Oct;38(10):751-757.

9. Ishioka P, Yamada S, Michalany NS, Hirata SH. Dermoscopy of Bowen's disease: pigmented variant on the penis. An Bras Dermatol 2012;87(3):482-484. Accessed 16 Jun 2022. Internet.

10. Carroll KA, Pierce J, Kovarik CL. Perianal Bowen disease in a child with human immunodeficiency virus. Pediatr Dermatol 2010;27(2):166-169.

11. Gahalaut P, Rastogi MK, Mishra N, Chauhan S. Multiple Pigmented Bowen's Disease: A Diagnostic and Therapeutic Dilemma. Case Rep Oncol Med 2012;2012:342030.

12. Park HE, Park JW, Kim YH, Han JH, Bang CH, Park YM, et al. Analysis on the Effectiveness and Characteristics of Treatment Modalities for Bowen's Disease: An Observational Study. J Clin Med 2022 May;11(10):2741-1. Accessed 22 Jan 2024. Internet.