JK SCIENCE pISSN: 0972-1177

CASE REPORT

Bowen Disease in Himachal Valley— An Under-Recognised Entity

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Abstract

Bowen's disease is under-recognised, in situ - squamous cell carcinoma usually presenting as a single, well demarcated, slow growing erythematous patch or plaque with a scaly or crusted surface. It has a limited potential of invasion.It mainly occurs over the sun-exposed parts of the body (typical variant). Genitals, back, finger, trunk & thigh are the peculiar sites for pigmented, verrucous, atrophic & hyperkeratotic variants which are considered as atypical variants. A total of 3 patients (2 males and 1 female; average age: 57 years) were noted over a span of one year. Rarity, atypical variants and limited case series on Bowen disease in literature prompt us to report this interesting case series.

Keywords:

Bowen Disease, Pigmented, Atypical, Verrucous, Sun Exposed Parts

Introduction

In 1912, John Bowen first explained this disease which is now termed as Bowen's disease.[1] Bowen's disease is under-recognised, in situ - squamous cell carcinoma usually presenting as a well demarcated, slow growing erythematous patch or plaque with a scaly or crusted surface. It has two variants clinically, typical and atypical. Typical variant mainly occurs over the sun-exposed parts of the body (head, neck and lower limbs). Atypical variants with respect to site include genitals, back finger, trunk & thigh. Morphological atypical variants include pigmented, verrucous, atrophic & hyperkeratotic types. Peak incidence seen in sixty to eighty years of age. It has a limited potential of invasion with an incidence of only 3%-5% in extra-genital & 10% in genital lesions.[2] Clinically, it occurs either as single or multiple lesions. Multiple lesions are seen mostly with arsenic exposure in 10%-20% of cases.[1,2] Pigmented BD is a rare variant of this disease which accounts for only 2% of all reported BD cases.[3,4,5] Here, we report a series of 3 cases of Bowen's disease with 2 atypical variants and 1 typical variant.

Case 1

A 60 year old male patient came to dermatology OPD with a single, slightly itchy, hyperpigmented to skin colored 3x3 cmverrucous lesion on posterior aspect of left thigh since 1 year. We received a punch biopsy measuring 0.5 x 0.5 x 0.3 cm in the histopathology department with differentials of verrrucous epidermal nevus, verruca vulgaris, squamous cell carcinoma, tubercular verruca cutis and Bowen Disease.

On microscopic examination epidermis showed marked hyperkeratosis, parakeratosis and purulent exudative

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Manuscript Received: 02.05.2024; **Revision Accepted**: 01.07.2024;

Published Online First: 10 Oct, 2024

Open Access at: https://journal.jkscience.org

Cite this article as: Puri M, Singh N, Gupta N. Bowen disease in Himachal valley-an under-recognised entity. JK Science 2024; 26(4):247-9

material. There was moderate nuclear pleomorphism in the keratinocytes with hyper-chromatic, dyskeratotic nuclei and increase mitosis with intact basement membrane. Upper dermis revealed marked lymphoplasmacytic infiltrate.

On account of above histopathological features the diagnosis of Bowen disease was given with exclusion of other differentials. On follow up, patient again had a relapse after 5 years.

Case 2

A 56 year old male presented to ENT OPD with a growth on left ary-epiglottic fold. We received two tiny tissue pieces measuring 0.1x0.1 cm in histopathology department. Haematoxylin and eosin stained sections showed hyperplasia and full thickness dysplasia of the epithelium. Atypical and bizarre mitosis along with chronic lympho-plasmacytic infiltrate also seen.

On account of the above histopathological features the diagnosis of Bowen disease was given. Comments upon invasion couldn't be given due to tiny biopsy. Patient was lost on follow up.

Case 3

A 60 year old Himachali woman came to dermatology OPD with a single well defined, hyper pigmented, hyper keratotic nodular lesion on the margins of left side perianal region. The lesion was associated with pain since three months. On palpation it was soft to firm in consistency. Excision biopsy was performed. A skin covered poly-poidal tissue piece measuring 0.6 x 0.6 x 0.2 cm was received in histopathology department with differentials of seborrhoic keratosis, viral wart, acrochordon, bowen disease and basal cell carcinoma.

On microscopic examination epidermis showed marked hyperkeratosis, parakeratosis, marked acanthosis, papillomatosis, focal areas of ulceration and bacterial colonies. Throughout epidermis there were loss of polarity, mild to moderate nuclear atypia, dyskeratosis, squamous pearls, individual cell keratinization, brisk mitosis and abundant melanin pigment.

Papillary dermis revealed dilated and congested blood vessels, increased melanophages and chronic inflammatory infiltrate. Reticular dermis was unremarkable.

On account of above histopathological features the diagnosis of Bowen disease (Pigmented variant) was given with exclusion of other differentials.

Discussion

Bowen disease is an in - situ squamous cell carcinoma which may affect either skin or mucosa or both. It has an incidence rate of approximately 1.42/1000 population.[6] Peak incidence is seen in sixty to eighty years of age but can occur at any age with no sex predilection. Solitary lesions mostly seen over sun exposed parts of the body (typical variant).[7] Multiple lesions are associated with arsenic exposure seen in 10%-20% of cases.Atypical variants with respect to site include genitals, back, finger, trunk & thigh.[7-9]

Our case series comprised of two patients with lesions on unusual sites (thigh,ary -epiglottic fold) and one patient with lesion at usual site (perianal region). Behera *et al* [8] and Sirka *et al* [7] reported two and one case presenting over thigh respectively. Similarly, Palaniappan and Karthikeyan [9] in their article on BD described a case over perianal region. No case have been reported so far in literature over ary-epiglottic fold.

Morphological atypical variants include pigmented, verrucous, atrophic & hyperkeratotic types.[10] In our case series, one case of verrucous type and one case of pigmented variant were noted which are very rare to find. Behera *et al* [8] reported a single case with verrucous morphology over thigh in his case series. We had atypicality in one of our case with respect to both site and morphology.

Pigmented BD is rare variant. Uncommonly it is found over the anogenital region of young and immunosuppressed adults. Melanocyte proliferation and melanin production in account of cytokines secreted by neoplastic cells can be the pathogenic mechanism. Due to increased function and size of melanocytes there is abundant melanin pigment in the cytoplasm of atypical keratinocytes, mainly in the basal layer.[11]

It can appear as a dark pigmented plaque with verrucous or hyperkeratotic surface. High risk strain of Human Papilloma Virus such as HPV 16 is usually responsible for occurrence in anogenital area. We don't have provision for virological study, here in our institute, that's why we cannot comment upon HPV association. [11, 12]

The etiology behind BD is damage to skin due to chronic exposure to UV radiation, arsenic exposure, immunosuppression, human papilloma virus infection, genetic factors and trauma.[5] However, there was no predisposing factor that can be correlated to any of our cases except HPV association. In all cases, routine

investigations including complete blood count, renal function test, liver function test, serology for HIV, hepatitis B and hepatitis C were within normal range.

Treatment : Topical 5 fluorouracil, topical imiquimod, topical diclofenac, cryotherapy, curettage, surgical excision, electrocautery, photodynamic therapy, ingenol mebutate (IM) and lasers.[13]

Conclusion

This case series presenting with typical and atypical variants of Bowen's disease highlights the diagnostic challenge for clinicians as it is a mimicker of common dermal conditions like eczema and psoriasis. Hope this case series bring recognition of BD in respect of its unusual presenting sites and morphology so that clinician can identify this entity early and prevent its misdiagnosis and malignant transformation. Regardless of its site, pigmented BD should always be considered in the differentials of pigmented skin lesions. Due to rarity of case series reported on BD in literature, we are presenting this case series. Clinical suspicion followed by histopathological examination are mandatory for its diagnosis.

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