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Case Report

Pigmented Bowen's disease over the right chest- A rare case report

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Abstract Pigmented Bowen's disease is one of the subtype of Bowen's disease which presents with a pigmented plaque arising in photo exposed areas of the body. We describe a case of Pigmented Bowen's disease in a 67-year-old male who presented with progressive growth of a pigmented and asymptomatic skin lesion of four years duration over right chest involving his right nipple. Dermascopy and histopathology was suggestive of Pigmented Bowen's disease. We report this case due to the pigmented variant which is a rare subtype and also the presence of lesion, over the sun-protected area, in an immunocompetent male in Indian type IV skin.

Key words
Dermascopy, Pigmented Bowen's disease, squamous cell carcinoma.

Introduction

Bowen's disease (BD) is a type of squamous cell carcinoma (SCC) in situ, which clinically presents as a well-demarcated erythematous scaly patch or solitary plaque with a crusted surface. Caucasians are the most commonly affected people with an incidence of about 1.42/1000.[1] Pigmented BD is a rare subtype of BD characterized by a hyperpigmented plaque, accounting for only 2% of all reported cases of BD.[2,3] Therapeutic options which are available include electrofulgration, cryotherapy, simple curettage, laser ablation, surgical excision, intralesional interferon alpha or bleomycin, photodynamic therapy and topical 5-fluorouracil.[4] Here, we present a case of pigmented Bowen's disease to raise awareness and challenging diagnosis of a rare variant of BD, in long standing pigmented lesions.

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Case report

A 67 year old, male reported to our dermatology department with complaints of progressive growth of a pigmented and asymptomatic lesion over the right chest involving his right nipple **(Figure 1)** of four years duration. On dermatological examination, well- defined hyperpigmented psoriasiform plaque was present over the right chest, measuring 5 cm in its largest diameter. Regional lymph nodes were not palpable. There was no history of any exposure to arsenic, radiation and there was no signs and symptoms suggestive of immunosuppression. General examination and other system examination were normal.

Complete blood count, renal function test and his liver function test were found to be normal. There were no abnormalities in his chest skiagram and ultrasound abdomen. With the above findings we considered the following differential diagnosis namely Seborrheic keratosis, pigmented basal cell carcinoma (BCC) and bowen's disease in this patient. Lesion was examined using a dermoscope DermLite DL3 (3Gen, Dana point, CA, USA) with 10X

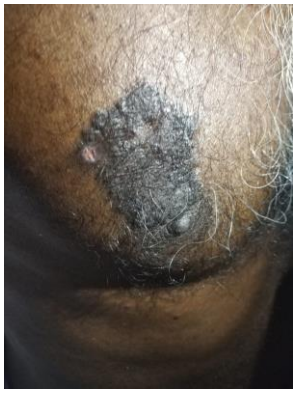

Figure 1 Hyperpigmented psoriasiform plaque present over right side of nipple.

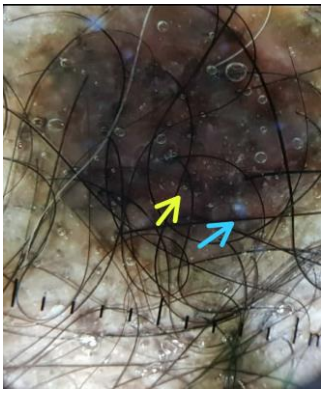

Figure 2 Dermascopy shows sharply defined edge with brown globules(yellow arrow) in patchy distribution with red dots and black globules with bluish hue (blue arrow) and few comedo openings.

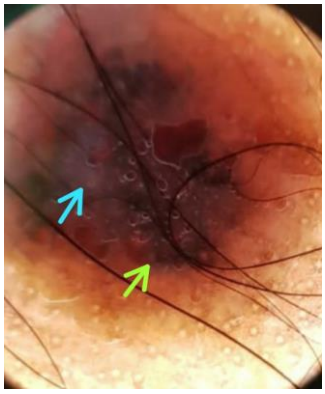

Figure 3 Dermascopy shows sharply defined edge (blue arrow) with gray-to-brown homogeneous pigmentation (green arrow).

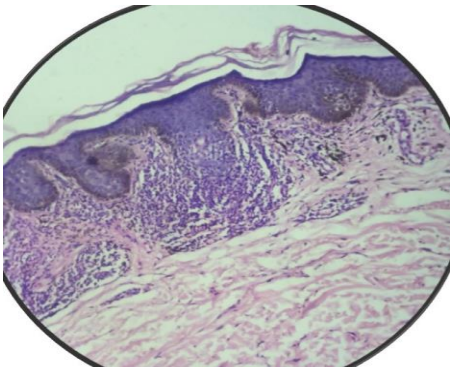

Figure 4 Photomicrograph showing Hyperkeratosis, acanthosis with intact basement membrane and irregular elongation of overlying epidermis into dermis with aggregation of inflammatory cells around epidermal extension. (H and E, x10).

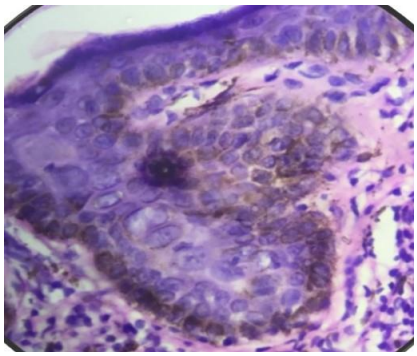

Figure 5 Photomicrograph showing atypical keratinocytes with intact basement membrane zone and aggregates of lymphocytes with melanin pigment incontinence and pigment laden cells.(H and E, x 40).

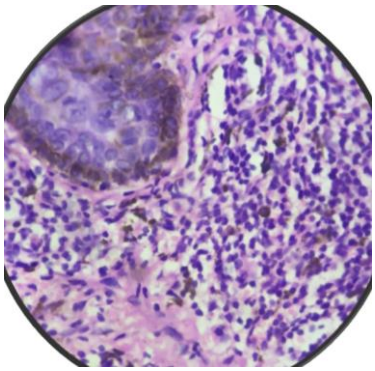

Figure 6 Photomicrograph showing pigment incontinence and lymphocytes aggregates with intact basement membrane zone. (H and E, x 40).

magnification equipped with an android phone in polarised mode with interface medium. On dermoscopy, we saw violaceous scaly plaque with sharply defined edge, brown globules in a patchy distribution with red dots and black globules with bluish hue and few comedo-like openings **(Figure 2, 3)**.

A skin biopsy was done with a 4mm punch from the right side of nipple lesion and sent to histopathology. The histopathological examination (HPE) showed parakeratosis, hyperkeratosis, acanthosis, full thickness atypia composed of medium to large sized atypical looking keratinocytes **(Figure 4)** with variable amount of cytoplasm, round to elongated hyperchromatic nuclei with moderate degree of nuclear pleomorphism. The cyptoplasm of atypical keratinocytes had melanin pigment, with intact basement membrane zone and aggregates of lymphocytes admixed with pigment laden cells **(Figure 5, 6)** in superficial dermis.

Absence of thread like raised borders, surface telangiectasia, peripheral palisading in HPE, arborizing vessels in dermascopy made us to exclude the diagnosis of pigmented BCC. The

diagnosis of Seborrheic keratosis was excluded because of the absence of stuck on appearance, horn cysts in HPE, milia like cysts in dermascopy. From the above dermascopy findings and HPE findings, we made a final diagnosis of Pigmented Bowen's disease. Topical diclofenac cream was prescribed and he did not turn up for follow up.

Discussion

John T. Bowen, a Boston dermatologist was the first to describe Bowen's disease (BD) in the year 1912.[5] BD is more common in persons older than 60 years of age but it may occur at any age.[6] Lower limbs, head and neck are the most common sites affected by BD. Irradiation (ultraviolet irradiation, radiotherapy, photochemotherapy), immunosuppression (e.g. after organ transplantation, AIDS), carcinogens (e.g. arsenic), viral (strong association of perianal and genital lesions with Human papilloma virus especially HPV 16), chronic injury have been reported as etiological factors for developing BD over several years.[7-10]

Pigmented BD is one of the rare subtype of Bowen's disease with a prevalence of 2% to 5% of all cases.[2,3] It clinically presents as a slow-growing, well-delineated, flat or slightly elevated, pigmented, variegated papule, or plaque with varying degrees of scaling that occurs in men mainly in their sixth to seventh decade, on the extremities (44%), followed by the trunk .[8,11]

One of the important feature of Pigmented BD is increased amount of melanin pigment in stratum basale, melanocytes and dermal melanophages.[12] The affected neoplastic cells may produce some specific cytokines which stimulate the production of melanosomes by melanocytes thereby increasing melanin synthesis.[12] It has been proposed by Satter that the pigmentation is because of the presence of an increased number of enlarged melanocytes with hypertrophic dendritic processes dispersed throughout the neoplasm.[13] Incidence of pigmented BD has been under reported in the studies which have been done in the past.[14] As dermoscopy is having higher sensitivity in detecting pigmentary changes, recent studies have argued a much higher incidence of pigmented BD, amounting to 38-64%.[15]

Glomerular and dot vessels grouped in clusters which are surrounded by a white halo (sign of keratinization) with surface scales are defined as typical dermoscopy findings of BD. These Glomerular vessels represents the dilated and tortuous vessels in the superficial papillary dermis which are easily seen with polarized mode of dermoscopy. Brown globules regularly packed in a patchy distribution along with gray-to-brown homogeneous pigmentation are the characteristic features of pigmented BD.[16] The optical interaction of polarized light with the skin around follicular infundibulae produces white rosettes which represents the interface between parakeratosis and orthokeratosis. Now a days, two more new features were described in the dermoscopy of BD namely double-edge sign (two parallel pigmented edges at the periphery of the lesion) and clusters of brown structureless areas.[17] These typical features has made dermascopy as one of the important tool in diagnosing BD. Other non-specific dermoscopic findings which can be seen in BD include comedo like openings, well-defined edges, structures in digital printing form and these findings are also seen in seborrheic keratosis.[18]

In any long standing pigmented lesions, pigmented BD should be considered as one of the diagnosis. Superficial basal cell carcinoma, bowenoid papulosis, seborrheic keratoses, pigmented actinic keratoses, melanocytic nevus, blue nevus, and superficial spreading melanoma were the differential diagnosis of pigmented BD.[9,10,19,20] Cryotherapy, electro cautery, photo

dynamic therapy, laser destruction, surgical excision, 5-fluorouracil cream, imiquimod cream and radiotherapy are the various therapeutic options available for the treatment of BD.[8]

Conclusion

To conclude, clinical suspicion is always required to diagnose atypical presentations of BD such as in this case. The presentation in our patient was unusual due to the pigmented variant, lesional location over a sun-protected area, in an immunocompetent male in Indian type IV skin. Pigmented BD should be in our mind as a differential diagnosis of any long standing pigmented skin lesion.

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