Giant pigmented Bowen's disease arising from seborrheic keratosis: morphology, dermoscopy, and surgical management with split-skin grafting

Jaspriya Sandhu, MD [1*]
Sunil Kumar Gupta, MD [1]
Shriya Garg, MBBS [1]
Sheerin Shah, MCh [2]
Bhavna Garg, MD [3]

1. Department of Dermatology, Venereology & Leprosy, DMCH, Ludhiana, Punjab, India
2. Department of Plastic Surgery & Burns, DMCH, Ludhiana, Punjab, India
3. Department of Pathology, DMCH, Ludhiana, Punjab, India
**Corresponding author:*
Jaspriya Sandhu, MD
Department of Dermatology, Venereology & Leprosy, DMCH, Ludhiana, Punjab, India
Email: sandhu.jaspriya@gmail.com

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Bowen's disease (BD) is an *in situ* squamous cell carcinoma of the skin. Pigmented Bowen's disease (pBD) is a rare variant, representing less than 2% of all BD cases. We report a case of Giant pBD arising from seborrheic keratosis in a 65-year-old man. The dermoscopy showed a scaly surface, structureless brown to grey-black areas with dotted vessels, glomerular and reticulate vessels, and a few brown globules. Histopathology was classical with a "windblown appearance"; prominent melanophages were seen in the dermis. Owing to the size and thickness of the plaque, a wide excision with a split-skin graft was done. The graft uptake was good, and the patient was followed up on days 4 and 26 post-surgery. We present our experience with the diagnosis and management of this rare case of giant pBD. To the best of our knowledge, such a case has not been previously reported.

Keywords: precancerous conditions, dermoscopy, Bowen's disease

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INTRODUCTION

Bowen's disease (BD), a form of in-situ squamous cell carcinoma (SCC), presents clinically as an erythematous plaque with fine scaling and crusting. BD is a pre-malignant condition; cumulative effects of ultraviolet radiation have a role in its etiopathogenesis [1]. The subsequent risk of developing an invasive carcinoma is approximately 3% [2].

Apart from its classical presentation, other clinical and histopathological variants of BD include acantholytic, papillated, pigmented, giant, dwarf, atrophic, hyperkeratotic, and pagetoid forms [1,3-7].

Pigmented Bowen's disease (pBD) is a rare variant characterized by increased melanin deposition in the epidermis or papillary dermis, seen commonly in elderly males. It presents as an asymptomatic, well-defined, pigmented plaque with fine scaling and crusting on non-sun exposed sites. The exact cause of the pigmentation is unclear, with pBD representing less than 2% of all cases of Bowen's disease [8,9]. The association of seborrheic keratosis with Bowen's disease has been reported in the literature; however, pBD arising from pigmented seborrheic keratoses is rare. We present a case of giant pBD with its clinical & dermoscopic features as well as the follow-up post-surgical excision and split-skin grafting.

CASE PRESENTATION

A 65-year-old male, farmer by occupation, presented with a pruritic dark skin lesion on the back, gradually enlarging over the past two years. On examination, a pigmented 4 x 5 cm plaque

was seen on the upper back over the left scapula (Figure 1a). The plaque was hyperpigmented to erythematous with well-defined, irregular borders, areas of thick crusting and fine scaling, and nodule formation at few sites within the plaque (Figure 1b). The back was also studded with multiple, discrete, pigmented papules and plaques of pigmented seborrheic keratosis (Figure 1a). There was no associated lymphadenopathy, organomegaly, or palpable abdominal mass. Dermoscopy of the lesion demonstrated a scaly surface with structureless brown to grey-black areas, dotted vessels, glomerular and reticulate vessels, and a few brown globules (Figures 2a–b) [DermLite© DL4W Dana Point, CA, USA].

A punch biopsy was taken, keeping the differential diagnoses of Bowen's disease, squamous cell carcinoma, basal cell carcinoma, and mycosis fungoides. Histopathological examination showed the presence of disorderly cell maturation seen as nucleomegaly and loss of polarity present within the entire thickness of the epidermis, giving it a *'windblown appearance.'* Atypical mitotic figures were also seen. The dermis showed chronic inflammatory infiltrate with prominent pigment incontinence. No areas of invasion were seen in serial sections (Figures 3a–b). On clinicopathological correlation, a final diagnosis of giant pBD was made, and surgical excision was planned. A wide excision with a 5 mm margin followed by split-skin grafting was done for the patient (Figure 4a). The patient was followed up on post-op days 4 and 26 (Figures 4b–c). There was good healing and graft uptake on follow-up. Multiple histopathological sections of the excised specimen did not show any areas of invasion into the dermis.

DISCUSSION

Pigmented BD (pBD) is a rare variant; in a series of 420 cases of BD by Ragi *et al.*, only 7 cases of pBD were seen [3]. Recently, Stewart *et al.* published

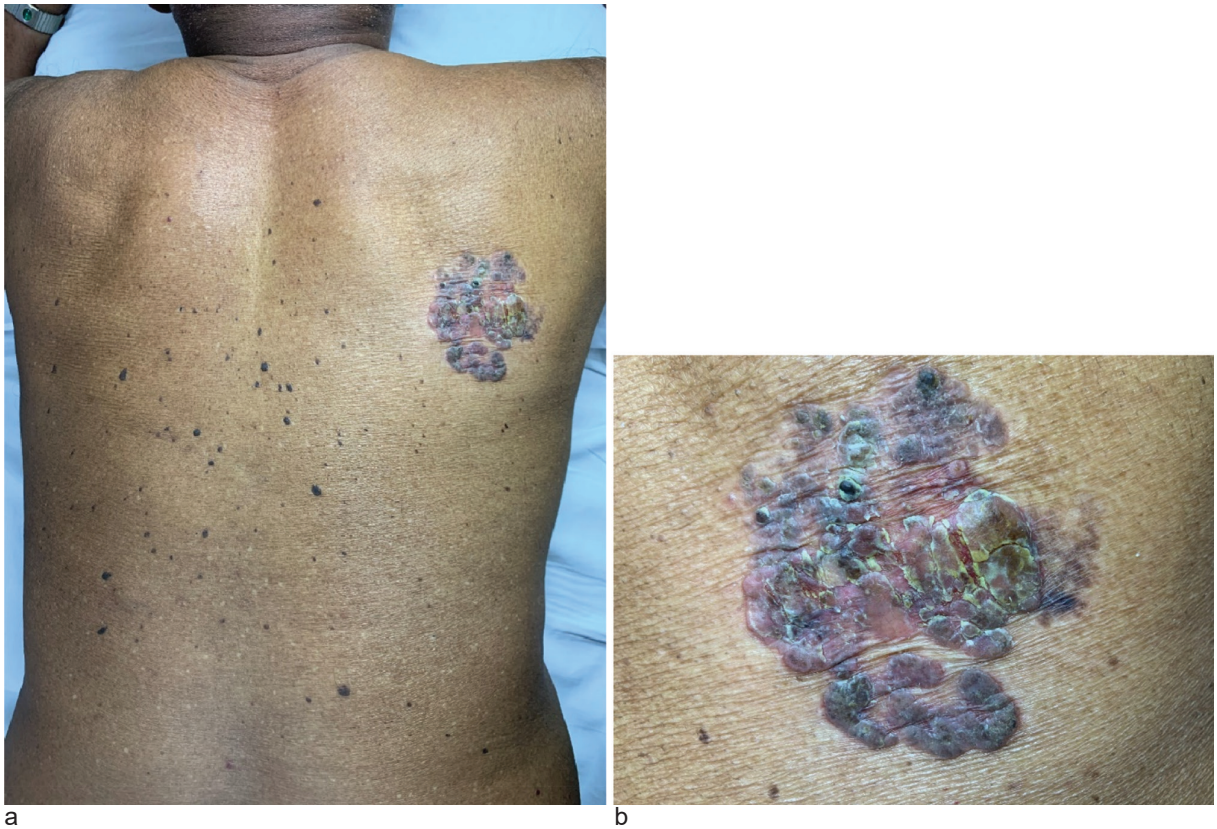

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Figure 1. (a) Back of the patient showing a large, well-defined, hyperpigmented plaque over the left scapular region. Multiple pigmented papules and plaques of seborrheic keratosis are also seen all over the back. (b) A hyperpigmented to erythematous, thick plaque with well well-defined, irregular borders; areas of thick crusting, fine scaling, and atrophy at a few sites within the plaque.

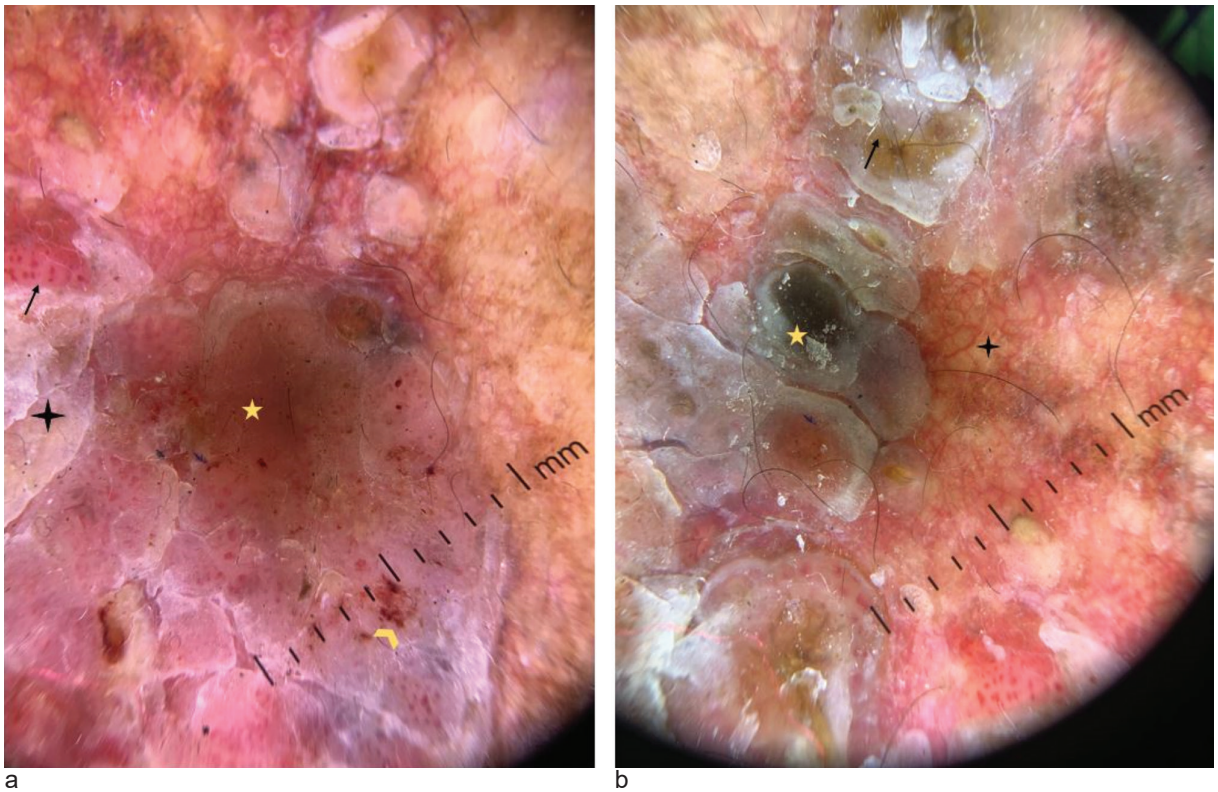

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Figure 2. Dermoscopy [10×, DermLite© DL4W Dana Point, CA, USA]: (a) Scaly surface (black star); structureless brown areas (yellow star); brown globules (yellow chevron arrow); dotted vessels (black arrow). (b) Thick scales (black arrow); homogenous brown to gray-black areas (yellow star); reticulate vessels (black star).

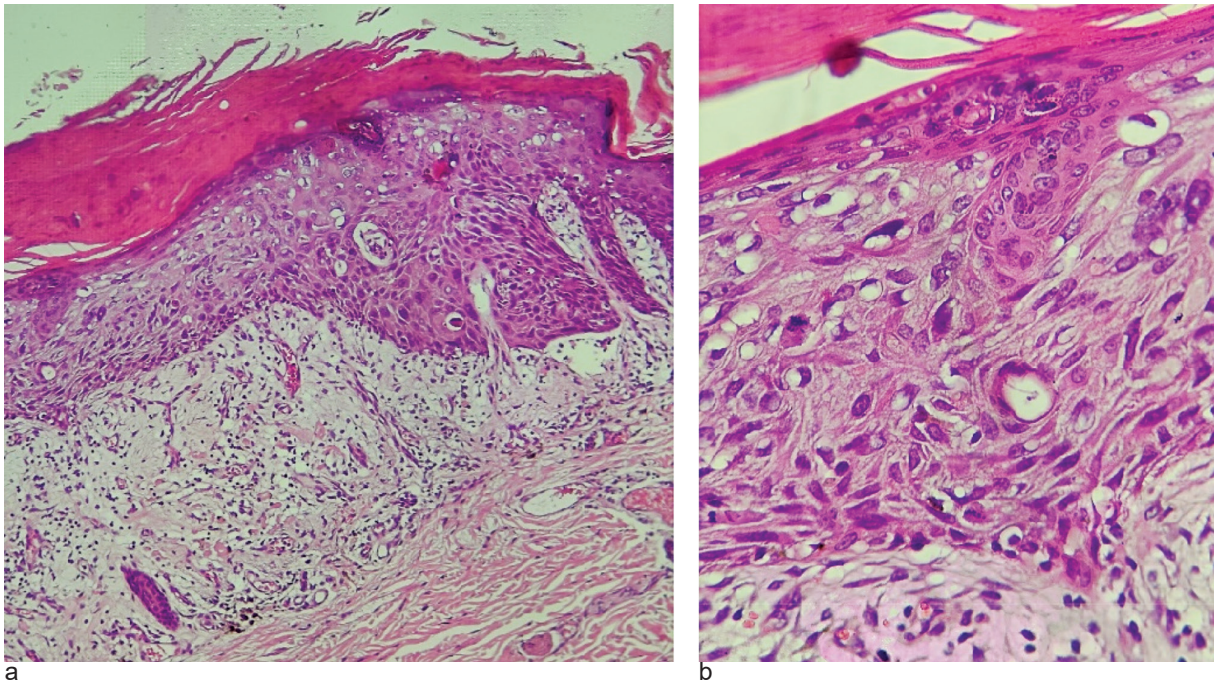

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Figure 3. (a) Hyperkeratosis; epidermis shows disorderly cell maturation. The dermis shows chronic inflammatory infiltrate with pigment incontinence (H & E, 100×). (b) Large cells with prominent nucleomegaly and hyperchromasia with atypical mitosis. Loss of polarity with a haphazard arrangement of cells, giving a characteristic "windblown appearance" (H & E, 400×).

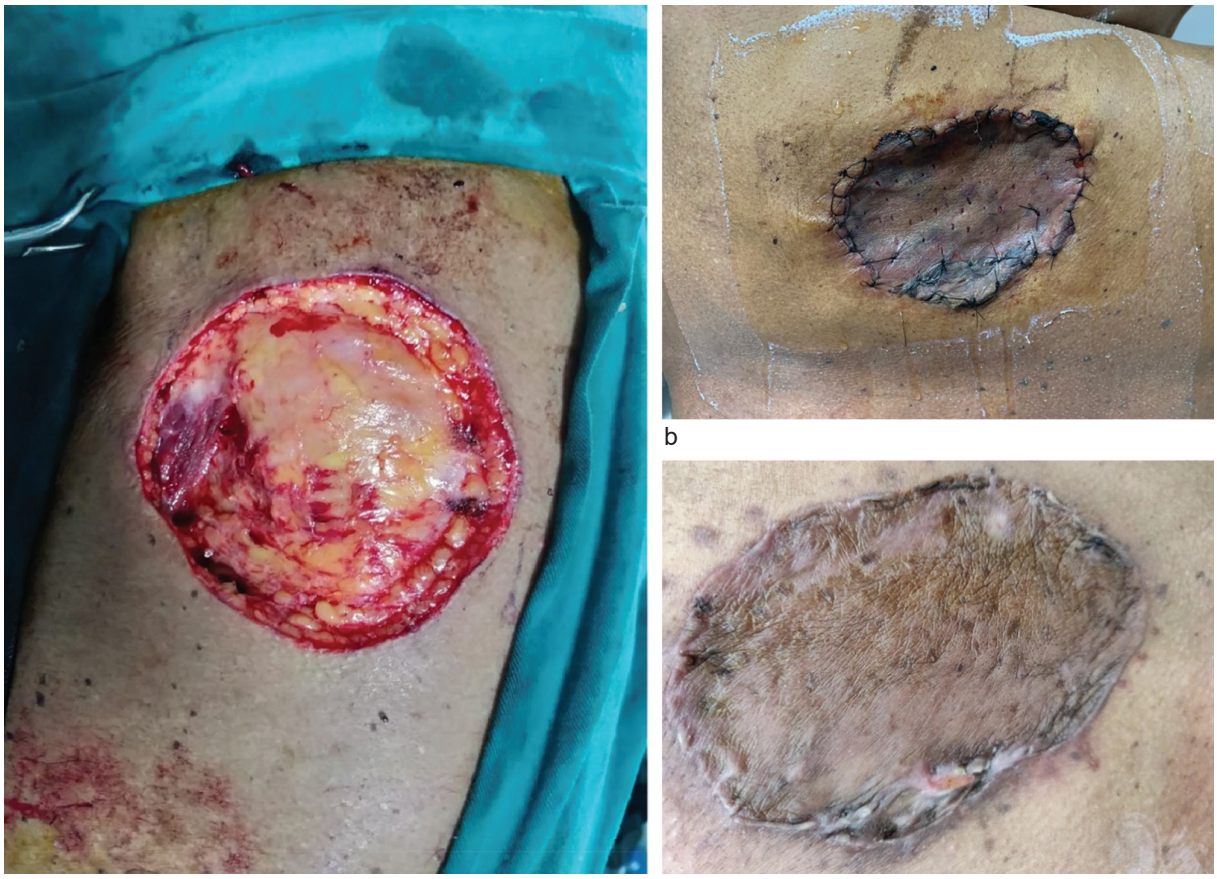

Figure 4. (a) Intra-operative image after excision with a 5-mm margin. (b) Post-operative follow-up on day 4 after excision and split-skin grafting. (c) Post-operative follow-up on day 26 after excision and split-skin grafting.

the largest review on pBD including 90 cases (out of 1,721 total BD cases) seen in an Australian dermatology clinic over a ten-year period. The mean age was 63.4 years, with a majority being male (64%) [10]. Similarly, our patient was a 65-year-old male.

Bowen's disease (BD) frequently arises in non-photo exposed areas, with factors other than sun exposure such as arsenic exposure, seborrheic keratosis (SK), and human papillomavirus infections having a role in the pathogenesis [11,12]. The transformation of SK to pBD was first reported by Bloch in 1978, while the transformation of SK to BD was reported by Christeler & Delaeretaz [11,13].

The clinical and dermoscopic features of pBD are not well described, and there may be common features with other skin malignancies. Histopathology remains the gold standard for diagnosis. The dermoscopy in a case of pBD on the penis was reported by Ishioka *et al.* and showed an amorphous light brown area adjacent to a dark brown area, containing irregularly distributed globules, streaks, and grayish-black portions [14]. Six cases of anogenital pBD with their clinical & dermoscopic features were described by Giuffrida *et al* [14]. They reported that glomerular vessels and linear grey/brown dots were the most important clearly visible dermoscopic clues for the diagnosis of pBD [15]. These are consistent with the findings seen in our case; however, these authors did not report giant pBD in their series [14,15].

Various treatment options are available for BD. These include topical modalities (e.g., 5-fluorouracil, diclofenac, imiquimod) and surgical modalities (e.g., cryotherapy, curettage with cautery, radiotherapy, laser, photodynamic therapy, and excision). The success of a treatment option depends on various factors like the site of the lesion, the number of

lesions, and the thickness of the lesion [12]. Owing to the large size & morphology (i.e., large, thick plaque), we opted for surgical excision, and the defect was repaired with a Thiersch graft (split-skin graft).

We report the case of an unusual, rare, giant pigmented variant of BD arising from seborrheic keratosis, with successful treatment & follow-up after surgical excision and split-skin grafting. To the best of our knowledge, such a case has not been previously reported.

Conflict of interest: None declared.

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