Our Dermatology Online

Case Report

Huge pigmented Bowen's disease: A rare case report

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ABSTRACT

Bowen's disease is an *in situ* squamous cell carcinoma with the potential to turn into invasive carcinoma. Giant Bowen's disease is its unusual variant with fewer reports, which poses a problem in the differential diagnosis with other pigmented lesions, especially when seen in sun-unexposed areas. It requires a joint effort of the dermatologist and surgeon to plan the line of management. Herein, we report a case of a seventy-year-old female patient with a large, multi-colored, asymmetric plaque with surface erosion in the left upper and outer gluteal region extending onto to the lateral thigh. Wide excision was done with a one-year follow-up showing no recurrence.

Key words: Bowen's disease, Squamous cell carcinoma, Rare variant, Wide excision

INTRODUCTION

Bowen's disease is one of the major histological types of non-melanoma skin cancer. In 1912, Professor John T. Bowen from Boston described this slow-growing premalignant cutaneous neoplasm as a form of *in situ* SCC with the potential to progress to invasive carcinoma [1]. Most studies have revealed a slight female preponderance [2]. Lesions are usually solitary, rarely multiple, mostly noticed in the head and neck region. However, the cheeks and lower legs are more likely to be affected in females [3]. They are usually asymptomatic, yet larger lesions may be pruritic and reach several centimeters in diameter. The giant form typically presents as a large, asymptomatic, reddish-brown plaque, scaly, with well-defined borders, and slowly growing. It affects any site, most frequently in sun-exposed areas. This intra-epidermal carcinoma is more common in the elderly. The pigmented/giant variant is relatively rare, with few reports in the literature. It poses difficulties in diagnosis and treatment. Histopathology is the gold standard diagnostic modality. Herein, we report a case of giant pigmented BD in a seventy-year-old female patient in a photo-protected site due to its rarity.

CASE REPORT

A seventy-year-old elderly female patient with no systemic diseases/immunosuppression presented with asymptomatic, reddish-brown discoloration of the skin with irregular borders with a variety of colors, approximately 7.5 cm in diameter, on the left upper and lateral buttocks extending onto lateral thighs for the past 11/2 years (Fig. 1). There was enlargement and darkening of the skin lesions for the past five months, which caused panic in the patient and relatives, who prompted her to seek treatment. There was no history of previous pre-existing skin diseases, cutaneous carcinomas, or arsenic exposure. Preoperative elliptical skin biopsy was taken and the H&E stained histopathology report revealed remarkable keratinocytes atypia with loss of cell polarity and the presence of some dyskeratotic cells, as well as an irregular and pronounced increase in melanin pigmentation in keratinocytes, both in the basal and in the scaly layer, characterizing pigmented giant BD (Figs. 2a and 2b). Owing to the mega-sized lesion, a wide excision was done (Figs. 3a and 3b). The margins were free of invasion by histopathology. The patient is regularly followed up (Fig. 4).

How to cite this article: Rajagopalan R. Huge pigmented Bowen's disease: A rare case report. Our Dermatol Online. 2025;16(1):66-68.

Submission: 11.04.2024; **Acceptance:** 04.10.2024

DOI: 10.7241/ourd.20251.12

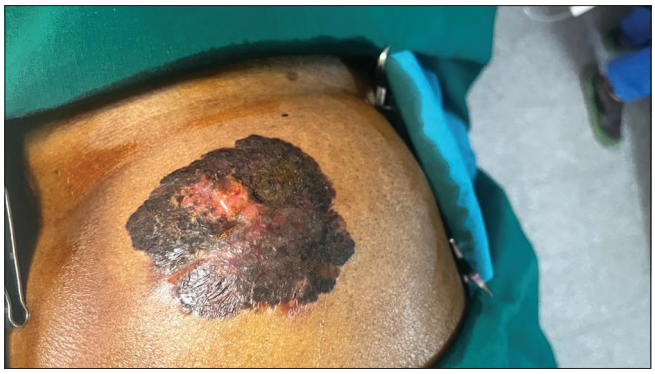

Figure 1: Giant Bowen's disease: A single, large, well-defined, multi-colored plaque.

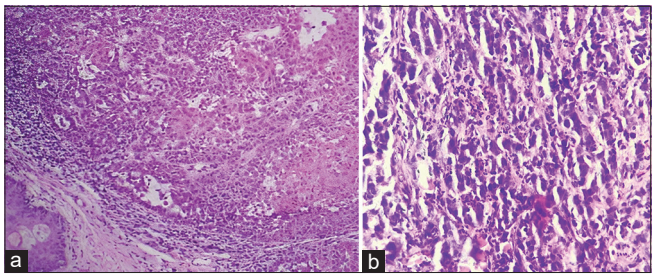

Figure 2: (a) Staining showing full thickness atypia (H&E; 10x). (b) Staining showing areas of hyperchromasia, pleomorphism, and atypical mitosis (H&E; 40x).

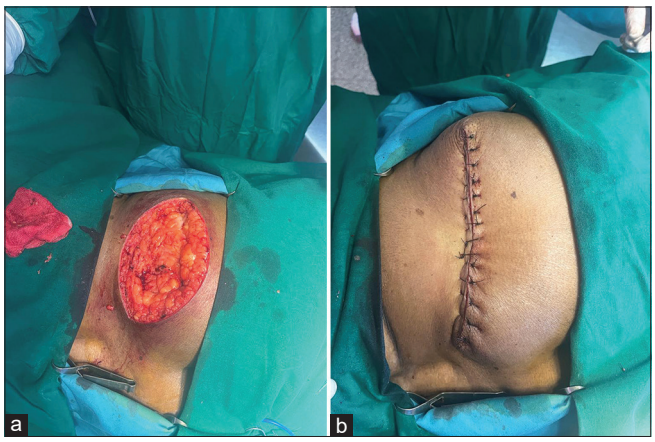

Figure 3: (a) Per-operative image. (b) The defect repaired by suturing.

DISCUSSION

BD typically occurs in persons above sixty years of age [4]. The giant pigmented BD is a rare variant that accounts for even fewer cases and is considered difficult to manage, necessitating tissue-sparing surgeries. Morton et al. defined "large Bowen's Disease" when the lesion dimension exceeds more than 2 cm [5]. Lopez et al. termed lesions with dimensions more than 3 cm as "extensive Bowen's Disease" [6]. It is characterized by the typical clinical picture of BD: full-thickness epidermal dysplasia/melanin increase in the epidermis

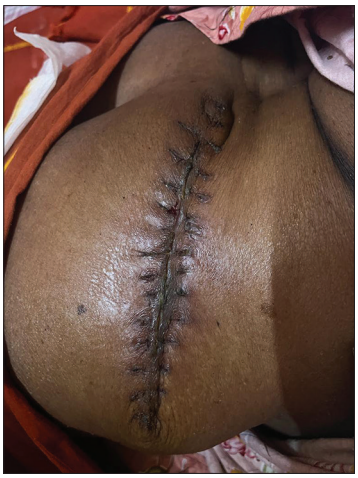

Figure 4: Healed skin site.

and dermis. It is clinically characterized by being large, multi-colored, hyperpigmented, well-defined, and with an uneven surface. Desquamation, erosion, and ulceration may be present, as well as itching or burning, although most lesions are asymptomatic. Risk factors are ultraviolet light exposure (solar light or phototherapy with UVA), organ transplantation, radiotherapy, immunosuppressive drugs, arsenic, thermal trauma, pre-existing skin diseases such as seborrheic keratosis, chronic lupus erythematosus, and lupus vulgaris and human papillomavirus (HPV) infection. Giant BD should be considered in the differential diagnosis (DD) of a huge, asymptomatic, pigmented plaque along with pigmented basal cell carcinoma, pigmented actinic keratosis, plaque psoriasis, and seborrheic keratosis.

Dermatoscopy assists in diagnosis, to exclude other causes. Findings noted are brownish, amorphous pigment (if any epidermal melanin) or grayish (if any dermal melanin), while brownish spots in a regular arrangement on the periphery of the lesion and dotted or glomerular vessels in a linear arrangement. These are not present in all cases. Our case had crops of rounded and coiled blood vessels apart from pigments. Histopathology is the gold standard for diagnosis. We found an acanthotic hyperpigmented epidermis with keratinocytes, showing atypia and loss of the usual pattern of maturation, an abundance of melanin in the cytoplasm of atypical keratinocytes, and a slight rise in the number of melanocytes, which confirmed our diagnosis. Although under- or late-diagnosed, the giant pigmented variant of BD may not be so rare. Challenges are in establishing the diagnosis early, and surgical treatment is the best option due to the large size, sometimes necessitating flaps. Risk-adapted follow-up is done regularly to look for local recurrences

and other keratinocyte cancers such as basal cell or squamous cell carcinoma elsewhere in the body [7,8].

CONCLUSION

The giant pigmented form is a rare variant of Bowen's disease, which may be seen in any keratinized skin. It carries an excellent prognosis if diagnosed early as it is a slow-growing pre-malignant condition. It requires a unified effort of a dermatosurgeon, one surgeon, and a plastic surgeon to plan and devise effective treatment options due to its varied presentation.

ACKNOWLEDGMENTS

We would like to acknowledge the patient for allowing me to use her images in the publication of this case report.

IEC approved the case report for publication.

Consent

The examination of the patient was conducted according to the principles of the Declaration of Helsinki.

The authors certify that they have obtained all appropriate patient consent forms, in which the patients gave their consent for images and other clinical information to be included in the journal. The patients understand that their names and initials will not be published and due effort will be made to conceal their identity, but that anonymity cannot be guaranteed.

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Source of Support: This article has no funding source.

Conflict of Interest: The authors have no conflict of interest to declare.