

Pigmented Bowen's disease: a case report of an unusual variant

Doença de Bowen pigmentada: relato de caso de uma variante rara

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ABSTRACT

Bowen's disease is a squamous cell carcinoma *in situ* with the potential to turn into invasive carcinoma. Pigmented Bowen's disease is an unusual variant, with few reported in the literature, and becomes a problem in the differential diagnosis with other pigmented tumors. We reported the case of a white female patient, 73-year-old, with a brownish asymmetric plaque, with more than one color, in the right upper limb, with 2-years of follow up.

Key words: squamous cell carcinoma; Bowen's disease; oncology.

INTRODUCTION

Bowen's disease (BD) is a squamous cell carcinoma *in situ* that is typically presented as asymptomatic erythematous plaque, scaly, well-defined borders, and slow growth^(1,2); it affects any area of the body, most frequently in sun-exposed areas⁽¹⁾. It is more prevalent in the elderly, affecting both sexes^(1,3). It has a rare pigmented variant, with few reports in the literature and difficult diagnosis⁽¹⁻⁵⁾. We report a case of pigmented BD in a 73-year-old female patient.

CASE REPORT

A 73-year-old white female patient, presenting brownish hyperpigmented plaque, asymmetrical, irregular borders, with more than one color, approximately 2.5 cm in diameter in the right upper limb, two years ago (**Figure 1**). There was growth and darkening of lesion in recent months. Personal history of squamous cell carcinoma (SCC) in anterior chest region excised two years ago. The lesion biopsy was performed, and histopathology revealed remarkable keratinocytes atypia with

loss of cell polarity and presence of some dyskeratotic cells, as well as irregular and pronounced increase in melanin pigmentation in keratinocytes, both in basal and in scaly layer, characterizing pigmented BD (**Figures 2 and 3**). Dermoscopy was not performed, because before medical follow up the patient died due to acute myocardial infarction.



FIGURE 1 – Hyperpigmented brownish plaque, asymmetrical, irregular border; more than one color, approximately 2.5 cm in diameter in the right upper limb



FIGURE 2 – Histopathology, 400× Fontana-Masson: Fontana Masson stain enhances melanin pigment. Melanin pigmentation is observed in the basal layer, focally in parakeratotic layer, and the presence of some melanophages in papillary dermis

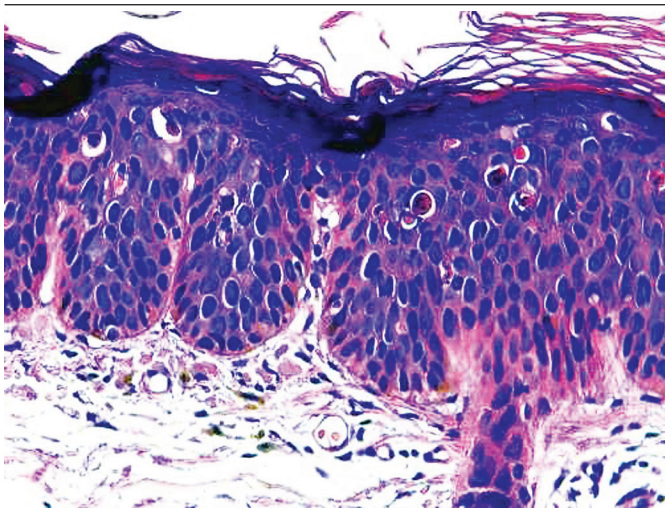


FIGURE 3 – Histopathology, 400× HE II: presence of dyskeratosis, atypical keratinocytes in full thickness of the epidermis, mitotic figures, pigment in the basal layer, and preserved epithelial basement membrane. The papillary dermis shows mild solar elastosis and melanophages

HE: hematoxylin-eosin.

DISCUSSION

The pigmented BD is a rare variant that accounts for less than 2% of BD cases, and is considered difficult to diagnose^(2,5). It is characterized by the typical alterations of BD and by melanin

increase in the epidermis and dermis^(1,2); clinically it appears as hyperpigmented, well defined, with a velvety surface, flat or verrucous plaque^(1, 2, 4). Desquamation and ulceration may be present, as well as itching or burning, although most lesions are asymptomatic^(2, 6). Risk factors are the same as BD's: exposure to sunlight, radiotherapy, arsenic, trauma, and infection with human papillomavirus (HPV)^(4, 6). Our patient had typical clinical lesion of slow growth and had a history of exposure to solar radiation, with SCC history.

Pigmented BD should be considered in the differential diagnosis of melanocytic lesions, pigmented basal cell carcinoma, pigmented actinic keratosis, seborrheic keratosis, and solar lentigo – often being associated with the last two^(7,9).

Dermatoscopy assists diagnosis, demonstrating the absence of criteria for melanocytic lesion, brownish amorphous pigment (if any epidermal melanin) or greyish (if any dermal melanin), while brownish spots in a regular arrangement on the periphery of lesion and dotted or glomerular vessels in linear arrangement are more specific^(7,9), but these features are not present in all cases; histopathology is the gold standard for diagnosis⁽⁷⁾.

By histopathology we found acanthotic epidermis with keratinocytes, showing atypia and loss of usual pattern of maturation⁽⁹⁾, abundant presence of melanin pigments in the cytoplasm of atypical keratinocytes and a slight increase in the number of melanocytes, which are typical^(2, 5-7). There is diffuse hyperpigmentation of epidermis, best observed in Fontana-Masson staining, melanophages in papillary dermis, and transepidermal elimination of melanin, which focally accumulates in the horny layer⁽⁹⁾.

Little is known about the mechanism of pigmentation of BD, and several theories attempt to explain its origin^(8, 9). The theory of “colonization” asserts that the pigment occurs due to the presence of an increased number of melanocytic hyperplasia with hypertrophic dendritic processes dispersed through the tumor⁽⁸⁾. Another theory suggests that, in pigmented BD case without scaling and keratosis signals, atypical keratinocytes are still in a well-differentiated stage, having, therefore, melanin⁽⁸⁾. In later stages, atypical keratinocytes lose this ability, resulting in loss of pigmentation, keratosis, and scaling. A third hypothesis suggests that specific growth factors or cytokines produced by cancer cells may stimulate the proliferation of melanocytes and melanin production^(8, 9). Also, the association and overlapping of pigmented BD with seborrheic keratosis and solar lentigo, some authors argue that in such cases, the preexisting hyperpigmentation spreads with the expansion of neoplastic

lesion and, as generally seborrheic keratoses and solar lentigo are not excised and subjected to histopathological routine examination, it is believed that the incidence of pigmented BD may be underestimated^(8,9).

The pigmented BD presents clinical similarity with other melanocytic lesions and should always be considered in the differential diagnosis of pigmented lesions. Although underdiagnosed, it may not be so rare, and its diagnosis is still a challenge.

RESUMO

A doença de Bowen é um carcinoma espinocelular in situ com potencial para transformar-se em invasivo. Ela é pigmentada, uma variante rara, possui poucos relatos na literatura e torna-se um problema no diagnóstico diferencial com outros tumores pigmentados. Relatamos o caso de uma paciente de 73 anos, branca, com uma placa acastanhada, assimétrica, com mais de uma cor, em membro superior direito, com evolução de dois anos.

Unitermos: carcinoma de células escamosas; doença de Bowen; oncologia.

REFERENCES

1. Plaza N, Kawa B, Dantas I, et al. Pigmented Bowen disease. *J Am Acad Dermatol.* 2005; 52(3): 148.
2. Firooz A, Farsi N, Firoozabadi MR, et al. Pigmented Bowen's disease of the finger mimicking malignant melanoma. *Arch Iranian Med.* 2007; 10: 255-7.
3. Lee JW, Hur J, Yeo KY, et al. A case of pigmented Bowen's disease. *Ann Dermatol.* 2009; 21(2): 197-9.
4. Hashemi P, Rabinovitz HS, Wang SQ. Confocal and histologic correlation of linear dots on dermoscopy of pigmented Bowen disease. *Am Acad Dermatol.* 2011; 64(2):77.
5. Ishioka P, Yamada S, Michalany NS, et al. Dermoscopy of Bowen's disease: pigmented variant on the penis. *An Bras Dermatol.* 2012; 87(3): 482-4.
6. Gahalaut P, Rastogi KM, Mishra N, et al. Multiple pigmented Bowen's disease: a diagnostic and therapeutic dilemma. *Case Rep Oncol Med.* 2012; 10(16): 1-5.
7. Cameron A, Rosendahl C, Tschandl P, et al. Dermatoscopy of pigmented Bowen's disease. *J Am Acad Dermatol.* 2010; 62(4): 597-604.
8. De Vries K, Lelie B, Habets WJ, et al. Pigmented Bowen's disease: a report of two cases. *Dermatol Surg.* 2011; 37(7): 1061-4.
9. Mota ANCM, Piñeiro-Maceira J, MFGS Alves, et al. A doença de Bowen pigmentada. *An Bras Dermatol.* 2014; 89(5): 825-7.

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