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CASE REPORT

Pigmented Bowen's disease presenting clinically as longitudinal melanonychia

Doença de Bowen pigmentada apresentando-se clinicamente como melanoníquia longitudinal

Lucas S. Madureira[1], Vivian W. Lederman[1], Julia C.K. El Dib[1], Paula F. M. Madureira[2], Lucas K. Fernandes[1], and Eduardo C.N. Constantino[3]*

[1]Department of Dermatology, Hospital de Base, Faculdade de Medicina de São José do Rio Preto; [2]Department of Dermatology, Faculdade Ceres; [3]Department of Pathology, Hospital de Base, Faculdade de Medicina de São José do Rio Preto. São Paulo, Brazil

Abstract

Bowen's disease (BD) is a squamous cell carcinoma, which affects more the elderly population and is more prevalent in light-skinned individuals. It affects most frequently sun-exposed areas and generally manifests as a single erythematous, scaly, or crusted plaque, with irregular growth and generally precise limits. Its pigmented variant is less frequent and the nail localization is even more atypical. The gold-standard treatment is surgical excision with safety margins. This report presents a case of pigment BD, in a female patient, on the fifth toe of the right foot, manifesting as longitudinal melanonychia (ML). The lesion had been present for a few years but there was recent darkening. The objective of the work is to raise awareness of the importance of BD as a differential diagnosis of longitudinal ML and to highlight the importance of histopathological studies for diagnostic confirmation.

Keywords: Bowen's disease. Squamous cell carcinoma. Epithelial and glandular. Neoplasms.

Resumo

A doença de Bowen (DB) é um carcinoma espinocelular (CEC), que acomete mais a população idosa e é mais prevalente em indivíduos de pele clara. Afeta mais frequentemente áreas expostas ao sol e geralmente se manifesta como uma placa única, eritematosa, escamosa ou crostosa, com crescimento irregular e limites geralmente precisos. Sua variante pigmentada é menos frequente e a localização ungueal é ainda mais atípica. O tratamento padrão-ouro é a excisão cirúrgica com margens de segurança. O presente relato apresenta um caso de Doença de Bowen pigmentada, em paciente do sexo feminino, no quinto dedo do pé direito, manifestando-se como melanoníquia longitudinal. A lesão já estava presente há alguns anos, mas havia escurecimento recente. O objetivo do trabalho é conscientizar sobre a importância do DB como diagnóstico diferencial da melanoníquia longitudinal e destacar a importância do estudo histopatológico para confirmação diagnóstica.

Palavras-chave: Doença de Bowen. Carcinoma de células escamosas. Neoplasias epiteliais e glandulares.

Correspondence:
Julia C.K. El Dib
E-mail: judib1998@gmail.com

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Introduction

Bowen's disease (BD) is a type of superficial intraepidermal squamous cell carcinoma (SCC), which becomes malignant in 3-5% of cases[1]. Its most common clinical topography comprises areas exposed to solar radiation, such as the head, neck, and extremities. Clinically, it is most typically characterized as a single, erythematous macule or papule, with a crusted or scaly surface, with well-defined edges and irregular growth. Risk factors for BD include female sex, white phenotype, human papillomavirus infection, history of arsenic exposure, and prolonged sun exposure. This condition tends to appear in the elderly, but the onset may be earlier in some cases[2].

The pigmented form of BD occurs in < 2% of cases[3]. It commonly presents as a hyperpigmented plaque (pinkish, brownish, and grayish-black) with a smooth, velvety, or hyperkeratotic surface, in intertriginous locations. Pigmented BD has an indolent, progressive course, and slow evolution, with patients being asymptomatic in most cases, but may also present local symptoms such as itching, bleeding, and pain, due to ulceration[4]. The pathophysiology of BD pigmentation is still not well understood. The most accepted theory relates to the accumulation of melanin in tumor cells and melanophages in the underlying dermis. However, this factor does not change the patient's prognosis and therapy[5]. The differential diagnosis of Bowen's disease is broad. It is often mistaken with psoriasis, multicentric superficial basal cell carcinoma, fungal skin infection (tinea cutis), nummular eczema, seborrheic keratosis and actinic keratosis, and actinic keratosis[6]. Specifically, pigmented BD can mimic malignant melanoma, seborrheic keratosis, pigmented actinic keratosis, solar lentigo, pigmented basal cell carcinoma, melanocytic nevus, and blue nevus[7]. When it affects the nail site, mimicking longitudinal melanonychia (ML), it appears as longitudinal bands a few millimeters thick, red, or black/brown and may develop hyperkeratotic or erythematous plaques around the nail[8].

Longitudinal ML most often has benign causes, such as physiological melanocytic activation, trauma, inflammation, nail nevus, and others[9]. Similarly, nail BD can also mimic several benign pathologies that occur with nail dystrophy, such as onychomycosis[2]. The malignant lesion of the SCC *in situ* type (BD) mimicking a longitudinal ML is unusual both due to the low frequency of its pigmented form (1.7% of cases) and its subungual location[10].

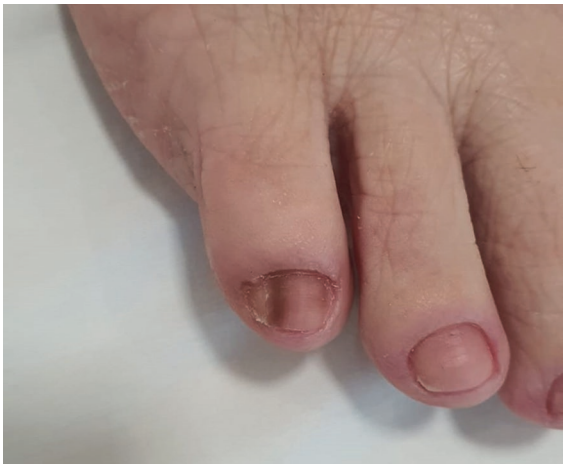

Figure 1. Longitudinal melanonychia of the nail plate, with irregular and heterogeneous pigmentation on the fifth toe of the right foot.

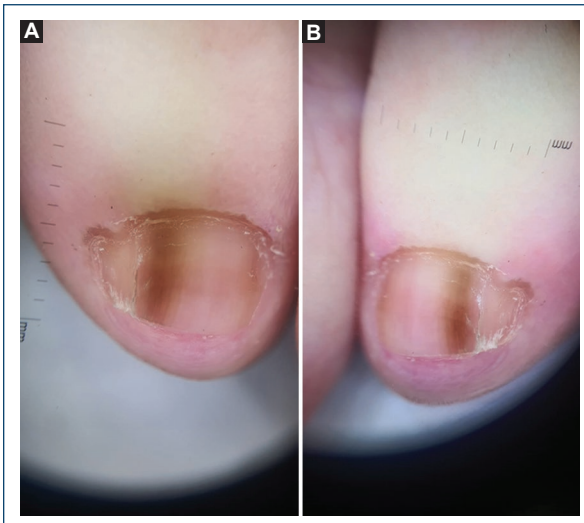

Figure 2. A and **B:** dermoscopy of the lesion with irregular light and dark brown longitudinal lines, approximately 3-mm wide.

Therefore, for a definitive diagnosis, in addition to clinical examination and dermoscopy, confirmatory histopathology is required[11].

Case report

A 43-year-old female patient sought a dermatologist due to a pigmented lesion extending longitudinally on the nail apparatus of the fifth toe of the right foot. A discrete subungual thickening and longitudinal

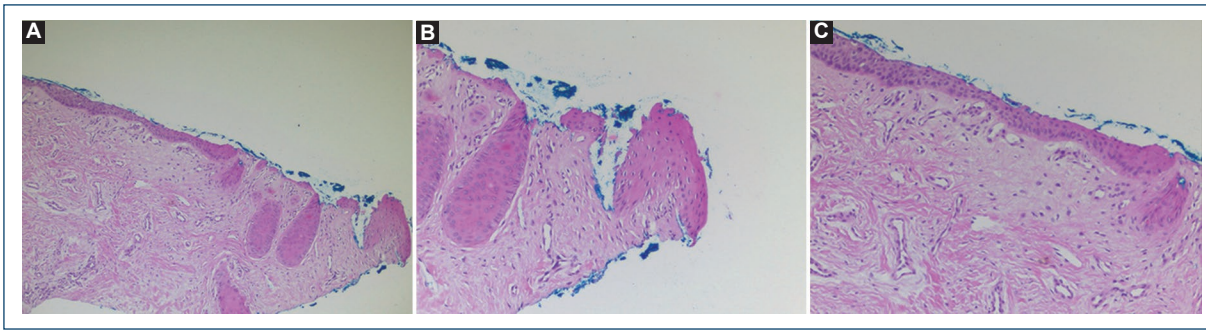

Figure 3. A-C: histopathology, showing hyperkeratosis and acanthosis with elongated and widened epidermal cones. Nuclear stacking and loss of epithelial polarity (H&E).

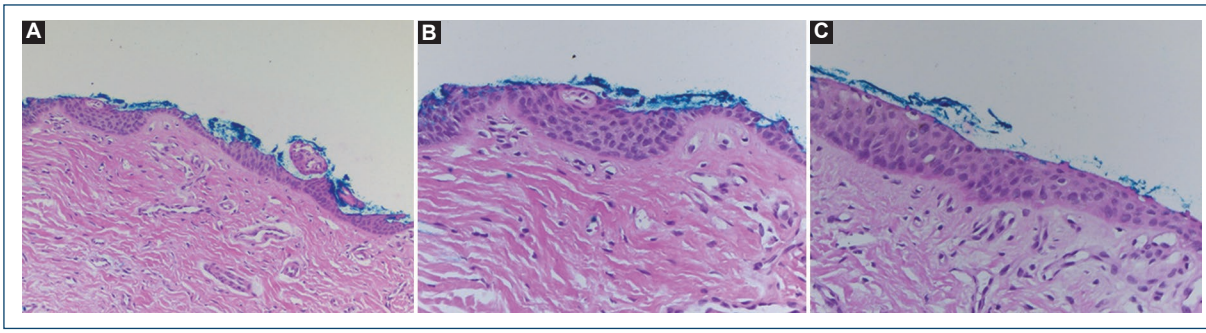

Figure 4. A-C: histopathological findings of lesion with proliferation of atypical keratinocytes and loss of polarity (H&E).

melanonychia (ML) with irregular pigmentation measuring approximately 3 mm in width were observed (Fig. 1). Dermoscopy of the nail plate showed irregular longitudinal lines of different colors, in shades of light and dark brown, as well as areas with hyperkeratosis (Fig. 2). According to the patient, the lesion had been present for a few years (the patient could not say exactly how long the lesion was present) but had recently darkened. The patient underwent an incisional biopsy of a nail matrix lesion on the fifth toe of the right foot. Histopathological examination demonstrated intraepidermal proliferation of atypical keratinocytes with loss of polarity (Fig. 3) and a nail plate with focal hyperkeratosis (Fig. 4). The findings revealed SCC *in situ* (BD), pigmented.

Discussion

Although SCC is the most common neoplasm of the nail system, nail BD has a variable frequency, possibly due to failure in recognition or underreporting. Therefore, pigmented BD of the nail apparatus is a rarely diagnosed condition and should be considered in the differential diagnosis of ML, distinguishing it mainly from nail melanoma. Typically, nail BD presents as subungual hyperkeratosis or a verrucous lesion of the nail plate or bed, with nail fold erythema and paronychia associated with crusts, ulcerations, or fissures; onychocryptosis and/or nail dystrophy; and rarely with ML.

This report draws attention to the fact that BD must always be remembered in the topography of the nail system, including in its unusual presentations. Histopathology examination remains the gold standard for confirmatory diagnosis, as in this case. However, careful assessment and management of patients with nail pigmentation, taking into account clinical features and dermoscopy, can help rule out common causes of nail pigmentation.

Conclusion

Thus, the present study highlights the importance of BD as a differential diagnosis of longitudinal melanonychia. Easy access to biopsies and histopathological studies allows early diagnostic confirmation of the

disease, which, combined with surgical treatment, guarantees the survival of the affected patient.

Funding

None.

Conflicts of interest

None.

Ethical considerations

Protection of humans and animals. The authors declare that no experiments involving humans or animals were conducted for this research.

Confidentiality, informed consent, and ethical approval. The authors have followed their institution's confidentiality protocols, obtained informed consent from patients, and received approval from the Ethics Committee. The SAGER guidelines were followed according to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing of this manuscript.

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