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Pigmented bowen disease on sclerosus and atrophicus lichen

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Abstract

Lichen Sclerosus and Atrophicus (LSA) is a chronic, inflammatory, mucocutaneous disorder of genital and extragenital skin. The malignant transformation had been described but rarely reported in the literature.

We report a particular case of pigmented Bowen's disease, which is a rare in situ form of squamous cell carcinoma, occurring on a sclerosus and atrophicus lichen.

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Introduction

Sclerosus or sclero-atrophic lichen is a fibrosing inflammatory dermatosis of chronic evolution and female predominance [1]. It manifested by a single leucoplasic, pigmented or erythroplasic plaque affecting, mainly the ano-genital region. Malignant degeneration is poorly reported in the literature. We report a particular case of pigmented Bowen's disease occurring on a sclerosus and atrophicus lichen.

Case presentation

A 41-year-old patient followed for a sclerosus and atrophicus lichen undergoing topical treatment without significant improvement. She had consulted in a urology department for dyspareunia with dysuria, and was referred for the management of an ulceration of the right labia majora. The clinical examination revealed an erythematous sclerotic cup of the genital area with effacement of the labia minora, as well as, a 2 cm ulceration of the right labia majora surrounded by a pigmented peripheral halo (Figure 1). Dermoscopy showed diffuse glomerular and linear vessels, homogeneous brown pigmentation in favor of Bowen's disease (Figure 2). Cutaneous biopsy confirmed this degeneration into Bowen's disease. The patient was referred to the gynecology department for surgical excision.

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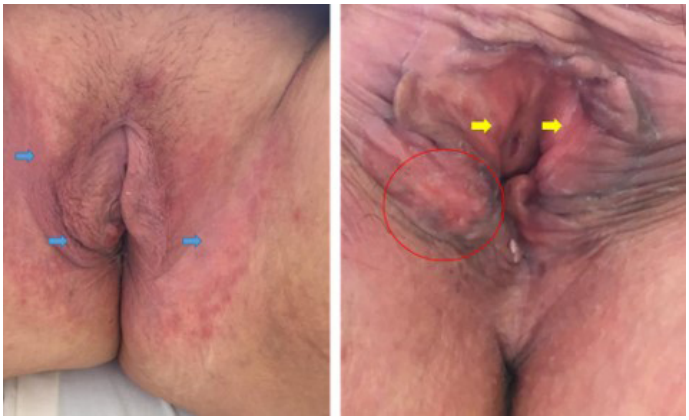

Figure 1: an erythematous sclerotic genital cup (blue arrows), effacement of the labia minora (yellow arrows) and a 2 cm ulceration of the right labia majora surrounded by a pigmented peripheral halo (red circle).

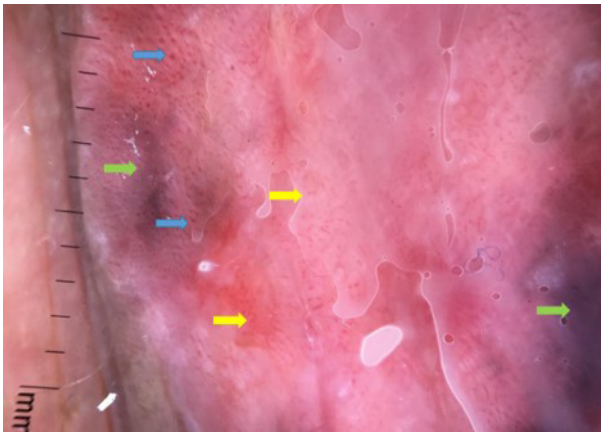

Figure 2: Dermoscopy showing glomerular (blue arrows), linear vessels (yellow arrows) and homogeneous brown pigmentation (green arrows).

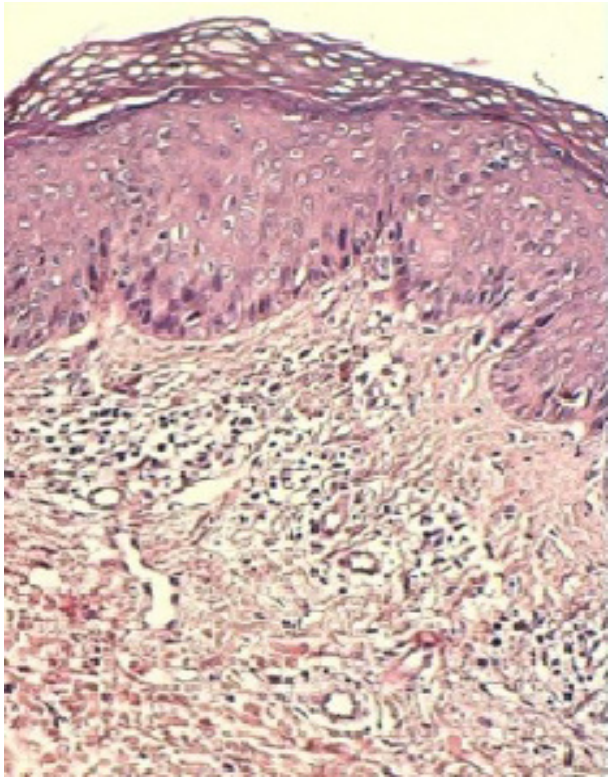

Figure 3: HES stain Gx200: parakeratotic hyperkeratosis and a cytoarchitectural disorganization with anarchic disposition of atypical keratinocytes and melanophages in the papillary dermis.

Discussion

Lichen Sclerosus and Atrophicus (LSA) is a chronic, inflammatory, mucocutaneous disorder of genital and extragenital skin. LSA is a debilitating disease, causing itch, pain, dysuria and restriction of micturition, dyspareunia, and significant sexual dysfunction in women and men [2]. It characterized by ivory-white plaques or patches with a glistening surface, which may become confluent extending around the vulval and perianal skin in a figure eight configuration [3]. The malignant transformation had been described but rarely reported in the literature. We describe this case because carcinoma had developed at an early age with a short course of the disease. Bowen's Disease (BD) is the in situ form of squamous cell carcinoma, often occurring in the chronically UV-damaged skin of elderly people [4]. BD is usually nonpigmented but it may also rarely be pigmented <2% of BD [4,5]. It is most frequent in women and occurs on the lower extremities in about three-quarters of patients [1]. BD typically presenting as a slowly enlarging, well demarcated erythematous to pink patch or plaque with irregular boarders and surface scale or crust [5]. That may be eroded or ulcerated. In contrast, pigmented BD is rare, and presents clinically as a nonuniformly pigmented plaque with a scaly or verrucous surface that should be differentiated from seborrhoeic keratosis, pigmented actinic keratosis, solar lentigo, basal cell carcinoma, blue naevus, melanocytic naevi and melanoma [4]. In our case, the recent appearance of a unique trailing ulceration was suspicious. A dermoscopy examination was performed showing specific findings of BD. A biopsy skin was essential to make the diagnosis and to role out other pathologies. Vascular structures (dotted vessels or 'glomerular' subtype morphology) plus a scaly surface are the most frequent dermoscopic finding of BD. In pigmented BD, small brown, black globules, homogenous pigmentation, pigmented streaks and pigmented network were supplementary features [5]. The histological finding of BD are full-thickness keratinocyte atypia, focal erosion, and occasional mitoses and dyskeratotic keratinocytes. Dense lymphocytic inflammation, dermal melanophages, and few scattered suprabasal dendritic melanocytes are seen in the heavily pigmented portion of the lesion [6]. Various treatment modalities are topical imiquimod cream, topical 5-fluorouracil cream, cryotherapy, surgical excision, curettage and electrocautery and photodynamic therapy, lasers and topical diclofenac [7]. In our observation, surgical excision was indicated because the lesion was accessible without undergoing an aesthetic or functional damage.

Conclusion

The risk of degeneration of sclero-atrophic lichen into Bowen's disease is strongly suspected in the presence of ulceration. All separate areas of ulceration must be biopsied in order to eliminate the possibility of a malignant transformation. For any patient with a sclerosus and trophicus lichen, rigorous monitoring is required to screen for malignant transformations.

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