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Bowen's Disease of the Vulva: A Rare Case and Review of the Literature

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1. Abstract

We present the case of a 27-year-old nulliparous woman with a history of cutaneous Rosai-Dorfman disease. She was previously treated with methotrexate and corticosteroids for this condition, and presented with six-year developing chronic lesions of the vulva. Histopathology showed Bowen's disease (squamous cell carcinoma in situ), which was HPV infection-associated. The patient was successfully treated with topical imiquimod therapy. This case highlights the importance of considering premalignant or malignant lesions in chronic vulvar dermatoses, particularly in immunosuppressed individuals.

2. Introduction

Bowen's disease or squamous cell carcinoma in situ is a rare but potentially precancerous disorder of full-thickness epithelial dysplasia without dermal invasion. Vulvar involvement is a minority of cases and is usually high-risk human papillomavirus (HPV) associated, particularly HPV-16 [1,2].Additional risk factors include chronic inflammation, lichen sclerosus and immunosuppression due to either systemic disease or therapeutic intervention [3,4]. In this context, we present a case study of vulvar Bowen's disease in a young woman with cutaneous Rosai-Dorfman disease who has been receiving long-term immunosuppressive treatment.

3. Case Presentation

A 27-year-old nulligravid woman who is known to have cutaneous Rosai-Dorfman disease and on long-standing corticosteroid and methotrexate therapy was referred from the dermatology department for chronic vulvar lesions for the past six years. She presented with a history of repeated but un-documented infections in the genital area.During physical examination, the patient was found to be in good general health. She was hemodynamically and respiratorily stable, normochromic conjunctivae, and had a BMI of 29 kg/m². Vulvar exam revealed dyschromic and hyperpigmented plaques (Figure1&2).The cervix was normal on speculum exam, no bleeding. Bimanual exam was of normal-sized uterus, no adnexal mass, and no palpable lymphadenopathy. Breast exam was normal. Targeted vulvar and adjacent skin lesions biopsies were performed. Histological analysis established Bowen's disease (squamous cell carcinoma in situ) in the background of HPV-induced epithelial alterations. Cytologic smear of the cervix showed an inflammatory background with atypical squamous cellscannot exclude high-grade lesion (ASC-H). Colposcopy was normal with type 1 transformation zone (TZ1), no acetowhite areas, and colpitis-like features.Screening for syphilis (TPHA), HIV, hepatitis B (HBsAg), and hepatitis C (anti-HCV) infections was negative.Topical imiquimod therapy was started in the patient. Clinical follow-up showed dramatic improvement of vulvar lesions.The patient was started on topical imiquimod therapy. Clinical follow-up showed significant improvement of vulvar lesions (Figure2).

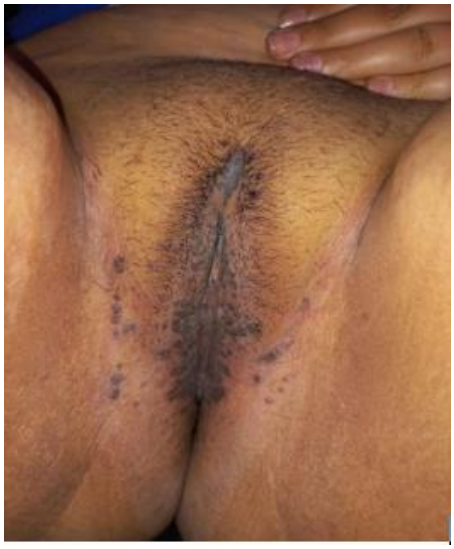

Figure 1: hyperpigmented, dyschromic vulvar lesions.

Figure 2: Evolution after imiquimod treatment.

4. Discussion

Vulvar Bowen's disease is a fairly uncommon localised squamous cell carcinoma in situ, particularly in young women. Known risk factors are high-risk human papillomavirus (HPV) infection, chronic immunosuppression, advanced age and dermatoses with chronic inflammation. In this patient, Rosai-Dorfman disease and immunosuppressive therapy probably led to HPV persistence and neoplastic transformation[5]. Recent evidence has implicated HPV, particularly high-risk types such as HPV-16, in the pathogenesis of vulvar intraepithelial neoplasia and Bowen's disease [6]. As there has been greater recognition of HPV-associated vulvar lesions in immunocompromised patients, there has been greater focus on diligent screening and early biopsy of suspicious lesions [7]. Topical therapies such as imiquimod have gained popularity as first-line or adjunct treatments in HPV-related intraepithelial neoplasia. Imiquimod is an immune response modifier with local cytokine release and antiviral effects. Its effectiveness in the management of vulvar Bowen's disease, particularly in those who wish to preserve vulvar anatomy and function, has been demonstrated in a number of case series and small studies [8].

Alternative therapies include surgical excision, laser, or photodynamic therapy, although these can cause scarring, anatomical distortion, and recurrence. Conservative options such as imiquimod are particularly valuable in young women and those with comorbidities [9].However, the potential for recurrence and long-term follow-up necessitate continued surveillance because recurrences can occur even after apparently successful treatment [10]. More recent development of HPV vaccination also brings up the potential for primary prevention of HPV-related vulvar lesions [11].Reports have emphasized that vulvar carcinoma in situ can be concurrent with, or progress to, invasive disease, particularly with late diagnosis or incomplete treatment [9].Furthermore, regular colposcopic monitoring has been recommended for those with multifocal HPV lesions by some reports.This case highlights the need for a multidisciplinary approach to chronic vulvar lesions, particularly in immunocompromised populations. Early diagnosis and treatment can prevent invasive cancer.

5. Conclusion

Immunocompromised women with chronic vulvar lesions require a complete workup, including biopsy. Premalignant conditions such as Bowen's disease can be identified early and treated conservatively and effectively. This case illustrates the necessity for careful management of longstanding vulvar dermatoses in young women who are at risk.

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