

Bowen's Disease of Unusual Localization

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Abstract

Bowen's disease is a carcinoma in situ. It occurs most often at an age of over 50 years, in light phototypes, in photo-exposed areas. Anal localization is exceptional. However, its association with condyloma is not accidental.

We report a case of perianal Bowen's disease occurring in systemic scleroderma.

Keywords: Bowen Disease; Systemic Scleroderma; Carcinoma

Abbreviations: BD: Bowen's Disease

Introduction

Bowen's disease (BD) is a carcinoma in situ described by Bowen in 1912. It occurs most frequently at an age of over 50 years, in light phototypes, in photo-exposed areas. Anal localization is exceptional.

We report the occurrence of anal Bowen's disease in a patient with systemic scleroderma.

Case report

A 48-year-old female patient, with sexual risk behaviours, followed for 6 years for systemic scleroderma according to ACR/EULAR 2013 criteria, with a non-fibrosing diffuse infiltrative lung disease for which she received 6 boluses of cyclophosphamide followed by azathioprine 100mg daily for 4 years with good clinical and spirometric evolution, was admitted with a clinical picture of chronic anal pruritus and alternating diarrhea and constipation, with no change in

general condition. Proctological examination revealed a perianal dyschromic infiltrate with cockscomb tumor lesions suggestive of condyloma (**figure1**). Histopathological examination of the placard biopsy showed an appearance compatible with Bowen's disease. Surgical excision was indicated at the multidisciplinary consultation meeting but refused by the patient.

Discussion

Bowen's disease is a squamous intraepithelial carcinoma. It is an in-situ form of cutaneous squamous cell carcinoma [1]. It is associated with cervical and vulvar intraepithelial neoplasia and have human papillomavirus as a common cause.

Its incidence is rare or underestimated. BD occurs in adults, with a peak in the seventh decade of life and a predilection for women.

Lesions may be single or multiple, and are ubiquitous, involving both photo-exposed and covered areas. Cutaneous involvement is the most common; mucosal or nail

localization is possible, but perianal localization is exceptional [2].



Figure 1: perianal dyschromic infiltrate with cockscomb tumor lesions

Patients with systemic scleroderma have a higher risk of cancer than the general population [3]. This risk particularly concerns solid bronchopulmonary and breast cancers [4], as well as hematological malignancies [5].

The pathophysiological mechanisms responsible for the increased prevalence of neoplasia in systemic scleroderma are manifold. Genetic [6], environmental and immunological factors, in particular the presence of anti-RNA polymerase III antibodies [7], and a history of renal crisis, have been described. Fibrosis in the various organs may also promote oncogenesis [5]. Finally, certain immunosuppressive treatments used in systemic scleroderma may be involved, such as cyclophosphamide [8].

Cyclophosphamide used in systemic scleroderma was associated with a higher incidence of cutaneous squamous cell carcinomas, angiosarcomas and urothelial carcinomas in situ in some studies [9].

Conclusion

Bowen's disease has a good prognosis. The risk of

progression to invasive carcinoma is 3-20%. BD is often treated with surgical excision.

Although several studies have highlighted the risk of associated cancer with systemic sclerodermia, long-term prospective data remain scarce, requiring clinicians to be vigilant in monitoring these patients.

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