A Case of Pigmented Verrucous Bowen's Disease in a Nigerian Woman

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ABSTRACT

Bowen's disease also known as squamous cell carcinoma (SCC) in situ is an uncommon premalignant lesion that affects both the skin and mucous membranes. Several risk factors have been identified including chronic exposure to ultraviolet radiation, immunosuppression, and exposure to some subtypes of human papilloma virus infection amongst others. The disease may be a differential for certain conditions like malignant melanoma and superficial basal cell carcinoma hence a histological diagnosis is essential in its diagnosis. Treatment options include topical, surgical and non-surgical ablative techniques. We report a case of Bowen's disease in a 61 year old Nigerian female trader who was managed successfully with topical 5 Fluorouracil cream.

Key words: Bowen's disease, squamous cell carcinoma in situ, risk factors, differential, treatment options.

INTRODUCTION

Bowen's disease also known as squamous cell carcinoma (SCC) in situ was first described by John T. Bowen, an American dermatologist in 1912[1]. This condition is a pre-malignant lesion that affects both the skin and mucous membranes and has the potential to become invasive SCC in about 3-5% of cases[2].

The etiology is largely multifactorial and implicating agents include; chronic exposure to ultraviolet (UV) radiation, arsenic exposure, human papilloma virus (HPV) particularly subtypes 16, immunosuppression, trauma, exposure to X-ray radiation and other chemical carcinogens.[1,3,4]

Bowen's disease occurs most frequently in sun exposed sites of Caucasians. It is rare in blacks with only about 43 cases said to have been recorded so far and tends to occur in non-sun exposed areas in this demographic[5, 6]. It has an almost equal sex distribution with a slight female preponderance[1]. It is a disease of adulthood, rare before the age of 30 years with the highest incidence in patients above 60 years[7].

There is paucity of data on both the worldwide and national prevalence of Bowen's disease. A study done in Minnesota in the United States in 1991 estimated the annual incidence to be 14.9 cases per 100,000 while another study from Hawaii in 1994 reported the annual incidence as 10 times that of the former[8,9]. In Nigeria two cases have been reported from Ahmadu Bello University Teaching Hospital Zaria, the most recent being in 2005 in a 59 year old Nigerian man[6].

Several clinical subtypes of Bowen's disease have been described including; erythematous, pigmented, hyperkeratotic, intertriginous and subungual or periungual subtypes[1].

We report a case of pigmented verrucous Bowen's disease in a 61 year old Nigerian woman. Malignant melanoma and pigmented superficial basal cell carcinoma were considered strongly as differentials, prior to histological confirmation of the diagnosis.

CASE REPORT

A 61 year old trader presented to the dermatology out-patient clinic of Lagos University Teaching Hospital (LUTH) with an 11 year history of a hyperpigmented painless lesion noticed on the lower 1/3rd of her left leg. There was no history of chronic exposure to X-ray irradiation, arsenic or repeated trauma. Her usual clothing consisted of long garments and her legs were always covered. She was not a known diabetic, hypertensive and was not on treatment for any chronic illness. She was not on long term use of steroids or any other immunosuppressive

medications. The lesion gradually expanded over the years with mild pain, which made the patient seek medical care.

Examination at presentation revealed a mildly tender hyperpigmented, verrucous, well circumscribed plaque, 5cm by 7cm, with slightly raised edges, on the anterior aspect of the lower 1/3rd of the left leg (Figure 1). Examination of other systems was essentially normal. HIV screening done was negative.

The histological examination of an elliptical skin biopsy revealed an intact stratum basalis, hyperkeratosis, parakeratosis and acanthosis of the epidermis with full thickness anaplasia also affecting the intra epidermal portion of the adnexia structures. There are numerous large atypical keratinocytes with abundant ground glass cytoplasm called 'pagetoid cells'. Based on the histologic findings, an assessment of Bowen's disease was made.

She was counseled on the diagnosis and available treatment options which include; Surgery, topical treatments with agents such as 5-Fluorouracil (5FU), Imiquimod cream. Patient opted for 5FU and was commenced on treatment-daily topical application with 1 fingertip unit covering the entire surface of the lesion. She responded to treatment with almost total flattening of the plaque after 12 weeks of treatment with 5FU but developed an ulcer within the improving lesion. (Figure 2) The 5FU was discontinued and site of lesion was treated with daily saline cleaning and mupirocin dressing. Patient was reviewed two weeks later and examination revealed a healed lesion with some post inflammatory hyperpigmentation. She is presently being followed up at the dermatology outpatient clinic.

DISCUSSION

Bowen's disease is rare in blacks, and requires a high index of suspicion to be diagnosed as it is most likely misdiagnosed as other skin conditions such as malignant melanoma. Patients frequently present with an asymptomatic, slowly enlarging well demarcated pink – erythematous scaly patch or plaque with irregular borders and overlying crust. The surface can be hyperkeratotic, verrucous or pigmented[1,5] as occurred with our patient who had the lesion for 11 years before presenting on account of new onset pain and increasing size.

Bowen's disease can affect both the skin and mucosal surfaces. It tends to occur more commonly as a single lesion on the head and neck in males and the lower extremities and cheeks in females[10]. Our patient typified this presentation with a lesion on the left lower limb.

Bowen's disease has various clinical subtypes as earlier mentioned and our patient presented with the pigmented subtype which is rare, occurring in less than 2% of cases simulating melanoma which was the initial working diagnosis prior to histological examination[1].

Bowen's disease has a 3 – 5% chance of progressing to invasive SCC, this is especially in lesions arising from the genitals (erythroplasia of Queyrat), one-third of which would be metastatic[1,2]. There is also a higher incidence of non-melanoma skin cancers in patients with Bowen's disease.

Clinically Bowen's disease is most often misdiagnosed as plaque psoriasis, melanoma, actinic keratosis, basal cell carcinoma, lichen planus, squamous cell carcinoma and inflamed seborrheic keratosis[11, 12]. Diagnosis is however confirmed by histologic examination of a skin biopsy which usually shows full thickness anaplasia of the epidermis including the intra epidermal portions of adnexal structures with complete distortion of the normal architecture. There is associated hyperkeratosis, parakeratosis and acanthosis. The stratum basalis remains intact, and there are numerous large atypical keratinocytes with abundant ground glass cytoplasm known as 'pagetoid cells'[1].

Treatment of Bowen's disease can be medical in form of topical therapies or surgical and non-surgical ablative techniques. The treatment of choice however depends on factors such as lesion site, size, number, available modalities and cost. Follow-up is highly essential because of the risk of recurrence. Factors affecting recurrence include a past history of recurrence, presence of multiple lesions, lesions in high risk locations and immunosuppression. Topical therapies include 5FU cream which was used in treatment of index patient and 5% Imiquimod cream. Surgical therapies include excision, Moh's micrographic surgery, curettage, chemo ablation and cryosurgery. Non-surgical ablative therapies include laser ablation, radiotherapy and photodynamic therapy.

5FU cream is easy to use and applied once or twice daily for about three months. Main adverse effects which our patient developed include skin irritation, ulceration and erosions that may last several weeks [13, 14]. Imiquimod cream is also used once daily for about 16 weeks and is associated with less skin irritation

than 5FU[15, 16, 17].

The prognosis of Bowen's disease is excellent; however there is a 13% risk of metastasis if Bowen's disease becomes invasive[1]. Frequent and routine screening for internal malignancy is also of utmost importance because of controversies associating Bowen's disease with internal malignancies[18,19,20,21].

In conclusion, Bowen's disease although rare, should be highly considered as a differential diagnosis in cases of slowly growing skin tumors resembling melanoma or superficial basal cell carcinoma and clinicians should also have a high index of suspicion. A histological diagnosis is pivotal in differentiating between these conditions.

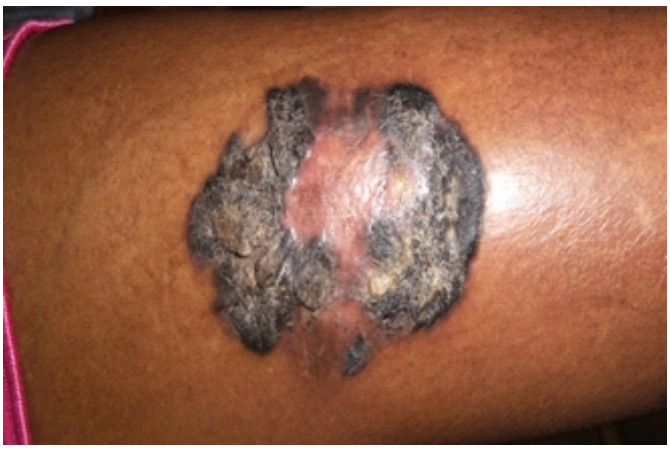

Figure 1: Lesion at presentation: hyperpigmented verrucous plaque on the anterior aspect of the lower 1/3rd of the left leg

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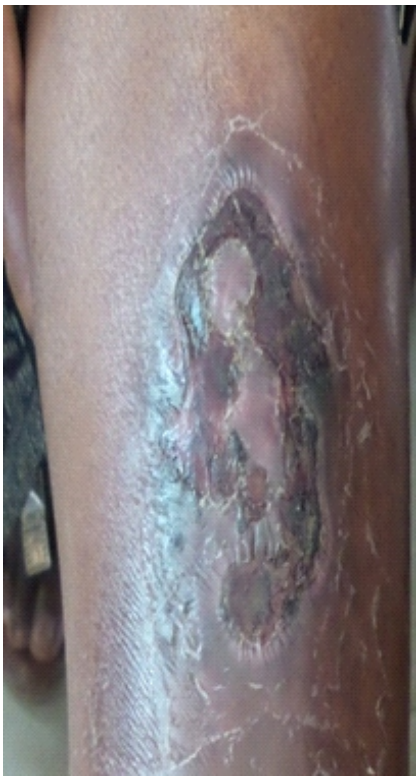

Figure 2: Lesion 12 weeks after treatment with 5-Fluorouracil

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