

A Disease in Disguise-A Case Report on an Atypical Presentation of Bowen's disease

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Abstract

Bowen's disease (BD) is a rare premalignant condition. The etiology of Bowen's disease is multifactorial. Classically, it presents as a well-demarcated erythematous plaque, predominantly in photo-exposed areas. The morphology of Bowen's disease differs based on the age of the lesion, the site of origin, and the degree of keratinization. Atypical variants in terms of morphology, such as verrucous, hyperkeratotic, atrophic, and pigmented types, are rarely described in the literature. Bowen's disease can be a diagnostic challenge for a physician as it mimics common conditions such as psoriasis, eczema, warts, etc. Histopathology is the gold standard diagnostic modality for confirming the diagnosis. The treatment depends on site, size, immune status, patient's age, aesthetic outcome, etc. The available therapeutic modalities include topical chemotherapy, surgical treatments, and light-based therapies. Here, we present a case report of an atypical presentation of Bowen's disease. A 68-year-old male came to the clinic with complaints of a raised skin-coloured lesion over his right ear for 45 days. On examination, a solitary, well-defined skin coloured – slightly yellow-coloured verrucous papule was seen over the helix of the right ear. A differential diagnosis of verruca vulgaris, Bowen's disease, and Bowenoid papulosis was made and sent for histopathology. Histopathological findings were consistent with Bowen's disease.

Keywords: Bowen's disease; Squamous cell carcinoma; Hyperkeratotic; Verrucous

Introduction

Bowen's disease is a rare, progressive intraepithelial carcinoma that was first described by John Bowen in 1912.¹ The risk of development of invasive squamous cell carcinoma is 3–5% in extra genital lesions and 10% in genital lesions.² It commonly occurs over sun-exposed areas such as the head, neck, and lower limbs. It usually presents as a solitary, well-defined erythematous plaque resembling psoriasis. But especially in cases of immunosuppression or chronic arsenicosis, multiple lesions may be seen. Uncommon clinical variants are verrucous, hyperkeratotic, and rarely pigmented ones. Here, we report a case of a 68-year-old male with an atypical presentation of Bowen's disease.

Case Report

A 68-year-old male presented with complaints of a painless skin-coloured raised lesion over his right ear for 45 days. There is a history of an abrupt onset and rapid progression of an asymptomatic, skin-coloured, rough, and raised lesion over the right ear. There was no history of prior trauma at the site of the lesion or any topical applications. There was no history of pain, itching, bleeding, or ulceration of the lesion. Systemic symptoms like fever, malaise, and weight loss were

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absent.

On clinical examination, a solitary, well-defined, non-tender, skin-coloured to slightly yellowish-coloured verrucous papule with crusting of 0.5x0.3x0.3mm was seen over the helix of the right ear (figure 1). Mucosae, hair, and nails were normal. No regional lymphadenopathy was noted. His systemic examination was unremarkable.

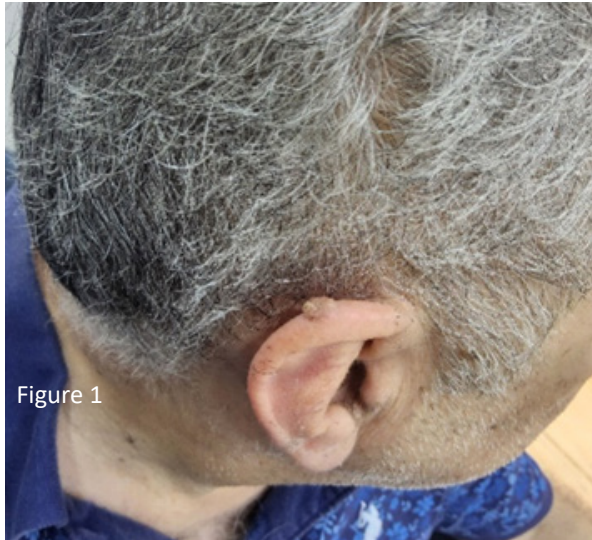


Figure 1

Figure 1: A solitary verrucous papule over the helix of right ear in a 68-year-old man

Routine investigations like complete blood count, liver function test, renal function test, and random blood sugars were within normal limits.

A differential diagnosis of verruca vulgaris, Bowen's disease, Bowenoid papulosis, seborrheic keratosis, and verrucous carcinoma was considered. An excisional biopsy of the verrucous lesion over the ear was done and sent for histopathological examination. HPE revealed a full-thickness epidermal dysplasia (figure 2A), with loss of polarity and a few abnormal mitoses (figure 2B) consistent with Bowen's disease (squamous cell carcinoma in situ). Eyeliner sign was seen in some places. There is a spread of disease laterally in the upper epidermis, leaving the basal layer intact. There was no evidence of dermal invasion. There was moderate (reactive) lymphocytic infiltrate in the papillary dermis. Complete excision was done during diagnosis. So the patient was asked to review once every 3 months to check for recurrence, if any.

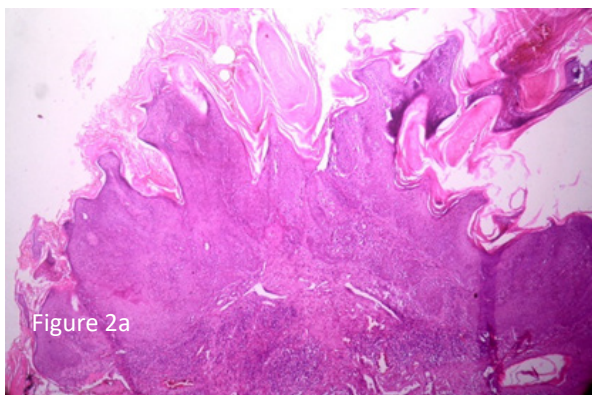


Figure 2a

Figure 2A: Haematoxylin and Eosin stain, 10x magnification showing complete architectural disarray of epidermis

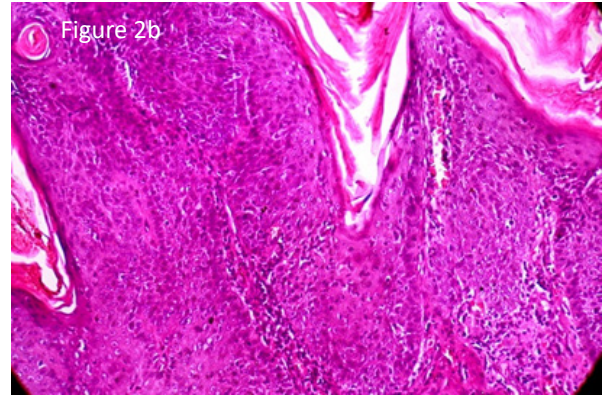


Figure 2B: Haematoxylin and Eosin stain, 40X magnification showing atypical keratinocytes with loss of polarity and abnormal mitoses

Discussion

Bowen's disease is a form of intraepidermal squamous cell carcinoma in situ with a small potential for invasive malignancy.¹ It may affect both skin and/or mucosa. The incidence is highest in Caucasians (1.42/1000 population).³ Bowen's disease usually affects fair-skinned individuals and is uncommon among type V Fitzpatrick skin people.^{4, 5} It can occur at any age but is mainly seen in the elderly, having equal sex preponderance. Here, we report a case of BD in a 68-year-old male of Fitzpatrick type.

BD occurs on skin damaged due to chronic ultraviolet radiation exposure, human papillomavirus infection, arsenic exposure, previous radiation, immunosuppression, trauma, and genetic factors.³ However, in our case, we did not find any predisposing factor to be correlated except intermittent moderate sun exposure.

BD usually occurs as a solitary lesion over sun-exposed areas.⁵ Multiple lesions are seen in 10–20% of cases with a history of arsenic exposure. The common sites for BD include the chronically photo-exposed sites such as the head, neck, dorsum of hands, and lower legs.^{5,6} Its occurrence in photo-covered sites is rare. The morphology of BD differs based on the age of the lesion, site of origin, and degree of keratinization.⁷ In keratinized areas, the lesions are erythematous and covered with scaling. In non-keratinized areas, they are velvety, smooth, and glistening. Morphology also varies in intertriginous and moist areas. Clinically, Bowen's disease presents as a solitary, well-demarcated, erythematous, crusted, or scaly plaque.¹ Other uncommon clinical variants include verrucous, hyperkeratotic, atrophic, and pigmented types.⁸ In the present case report, there is an atypical solitary skin-coloured to yellow-coloured verrucous papule seen over the helix of the right ear. Bowen's disease presents as a verrucous papule in our case, mimicking

verruca vulgaris is rare.

The diagnosis of BD is based on a high index of clinical suspicion, which is supported further by histopathology. Histological examination of Bowen's disease reveals a hyperkeratotic epidermis with a variable degree of parakeratosis. The cells throughout the epidermis lie in complete disorder, resulting in a "windblown appearance" showing atypia with large hyperchromatic nuclei with intact basement membrane.¹ In our case, we found cellular disarray and atypia with an intact basement membrane, indicating that there was no progression towards squamous cell carcinoma.

Various treatment modalities are used, including topical imiquimod cream, topical 5-fluorouracil cream, topical diclofenac, surgical excision, curettage, electrocautery, cryotherapy, photodynamic therapy, and lasers.⁹ Here, we successfully treated the condition with laser excision.

Bowen's disease usually has an excellent prognosis because it is a slow-growing premalignant lesion. Even spontaneous regression of BD has been reported, probably due to Fas-mediated apoptosis.¹⁰ In BD, recurrence is relatively rare and is approximately 6% within 5 years of taking sufficient treatment. The

recurrence is more common among immunosuppressed individuals.¹¹

We are reporting this case because only a few case reports have been described with this unusual novel presentation as verrucous papules are less reported and easily misdiagnosed. Immunosuppression and arsenic exposure are not risk factors in every case of Bowen's disease.

The findings of this case report are not representative of typical patient outcomes, and the statistical analysis is not feasible, limiting the ability to draw robust conclusions about the significance of the findings.

Conclusion

The asymptomatic nature, slow progression, and non-specific, varied presentations of Bowen's disease make the clinical diagnosis challenging even for experienced dermatologists. Considering the risk of malignant transformation, a high index of suspicion, supported by histopathology, is required to diagnose and treat this condition without delay, which in turn may reduce the morbidity of patients.

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