Bowen's disease on two different unrelated anatomical sites (genitals and nail) in succession in an immunocompromised patient

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Abstract

Bowen's disease (BD) is a premalignant condition. Its exact etiology is unknown but chronic arsenic and sun exposure, and human papillomavirus infection is known predisposing factors. Pigmented lesions of BD represent 1.7%–5.5% of all BD cases. BD in the nail unit is challenging due to its varied clinical presentations such as fissure, ulceration, warty lesion, paronychia, onychocryptosis, and nail dystrophy. We present the case of a 43-year-old married, immunocompromised male (HIV), with a CD 4 count of 478, on tenofovir, atazanavir boosted with ritonavir regimen, known diabetic presented with multiple asymptomatic discrete, rounded, hyperpigmented verrucous papules on both surfaces of shaft of penis and scrotum and a single, 4 cm × 3 cm, irregular, smooth surfaced, hyperpigmented plaque, on the base of the penis extending to the upper part of the scrotum of 1-year duration with history of multiple unprotected sexual exposures with unknown female partners. Regional lymphadenopathy and systemic complaints were absent. Biopsy from hyperpigmented verrucous papule and hyperpigmented plaque was consistent with verruca vulgaris and pigmented Bowen's disease, respectively. The patient was lost to follow-up. Ten months later, he presented with longitudinal melanonychia with a subungual hyperpigmented mass protruding beyond the distal nail margin near the lateral nail fold of the right middle finger nail with an absent Hutchinson's sign. Longitudinal excisional biopsy of nail lesion was consistent with BD. He was started on 5-fluorouracil 5% for BD of genitals and podophyllin application for verruca vulgaris with remarkable improvement in both the lesions and there is no recurrence of nail lesion after 9 months of excision.

Key words: Bowen's disease, longitudinal melanonychia, pigmented variant

Introduction

Bowen's disease (BD) is an intraepidermal squamous cell carcinoma (SCC) *in situ* affecting both sun-exposed and nonsun-exposed areas. It is a premalignant condition. The pigmented variant of BD is unusual accounting for only 1.7%–5.5% of all cases.[1] Exact etiopathogenesis is not known but certain predisposing factors have been implicated. BD of the nail unit is rare and diagnosis is usually difficult due to its varied clinical presentations. The explanation for the coexistence of the BD of genitals and nail unit is based on anogenital-digital spread theory.[2]

Case report

A 43-year-old married, immunocompromised male, with a CD 4 count of 478, on tenofovir plus atazanavir boosted with ritonavir regime, known diabetic on insulin, oral hypoglycemic presented with multiple asymptomatic raised lesions on the shaft of the penis and scrotum and single flat black lesion on the base of the penis for 1 year. He gave a history of multiple unprotected sexual exposures with unknown female partners. He had no systemic complaints. There was no history suggestive of arsenic exposure.

Cutaneous examination revealed multiple, discrete, rounded, hyperpigmented verrucous papules, ranging from 3 to 5 mm on dorsal and ventral surfaces of the shaft of the penis [Figure 1a and b] and a single, 4 cm × 3 cm, irregular, smooth surfaced, hyperpigmented plaque, on the base of the penis extending to the upper part of the scrotum [Figure 1c]. There was no regional lymphadenopathy. Hemogram, chemistry profile, and radiological studies were normal.

Histopathology from the plaque showed an acanthotic epidermis with atypical keratinocytes spanning the full thickness of the epidermis giving rise to a windblown appearance with intact basement membrane and dermal melanophages, findings consistent with pigmented

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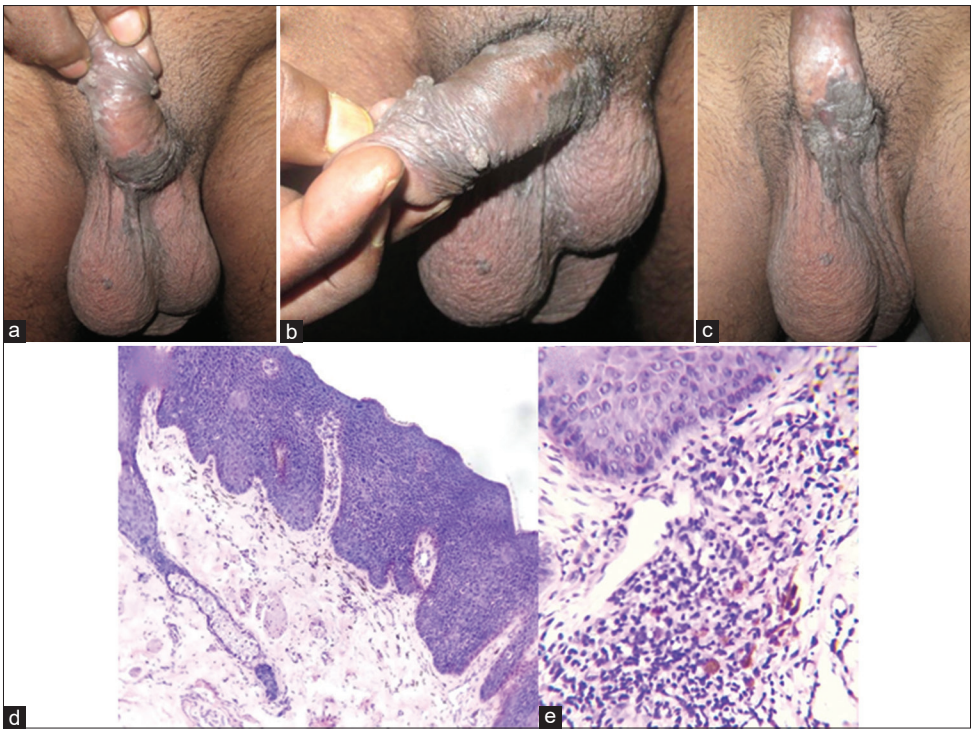

Figure 1: (a and b) Multiple, discrete, rounded, hyperpigmented verrucous papules, ranging from 3 to 5 mm on dorsal and ventral surfaces of the shaft of the penis. (c) Single, 4 cm × 3 cm, irregular, smooth surfaced, hyperpigmented plaque, on the base of the penis extending to the upper part of the scrotum. (d) (H and E, ×40): Biopsy from hyperpigmented plaque showed acanthotic epidermis with atypical keratinocytes spanning the full thickness of the epidermis giving rise to a windblown appearance with intact basement membrane and dermal melanophages. (e) (H and E, ×400): Biopsy from hyperpigmented plaque showed atypical keratinocytes with intact basement membrane and dermal melanophages

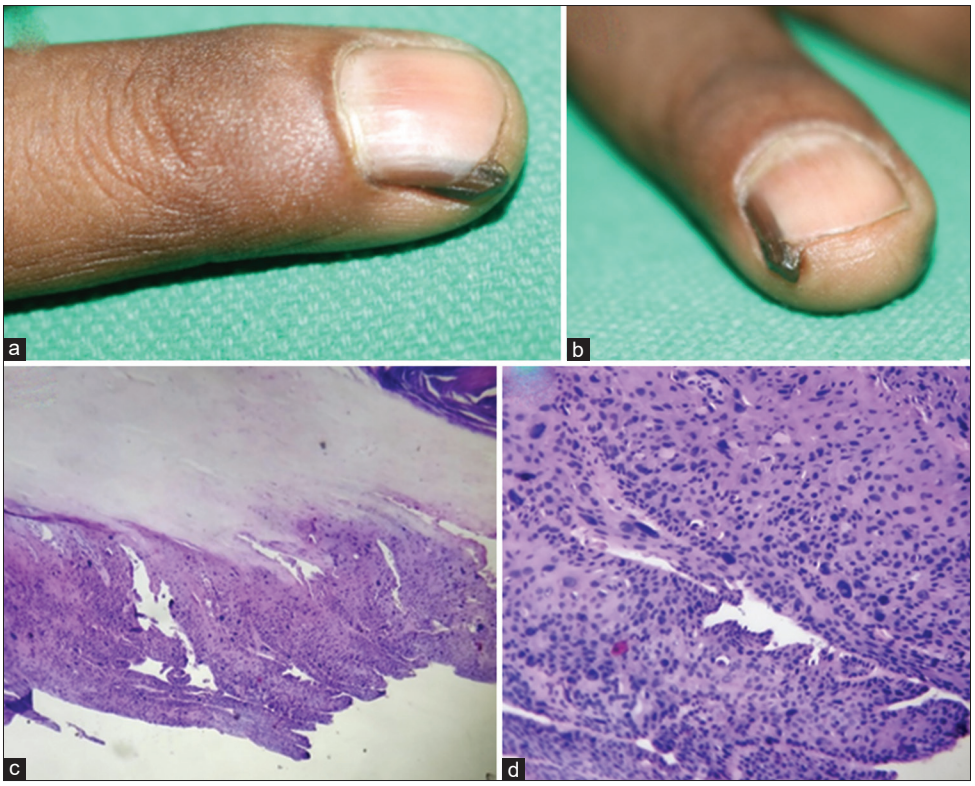

Figure 2: (a and b) The right middle finger nail showed a longitudinal melanonychia involving < 1/4th of the nail with a subungual hyperpigmented mass protruding beyond the distal nail margin. (c) (H and E, ×40): Longitudinal excisional biopsy of nail lesion showed thickened nail plate, acanthotic epidermis, and atypical keratinocytes spanning the entire epidermis. (d) (H and E, ×400): Longitudinal excisional biopsy of nail lesion showed atypical keratinocytes

BD [Figure 1d and e]. Biopsy from the verrucous papule showed hyperkeratotic stratum corneum, acanthotic epidermis with papillomatosis, and confluent rete ridges with koilocytes suggestive of verruca vulgaris.

Treatment planned was 5-fluorouracil 5% for the BD and podophyllin application for verruca vulgaris, but he was lost to follow-up.

Ten months later, he presented with blackish discoloration of the lateral aspect of the right middle finger nail with blackish subungual mass projecting beyond the distal nail margin. He reported no preceding trauma. There was the persistence of the verruca vulgaris and BD.

The right middle finger nail showed a longitudinal melanonychia involving <1/4th of the nail with a subungual hyperpigmented mass protruding beyond the distal nail margin [Figure 2a and b]. Hutchinson's sign was absent. Longitudinal excisional biopsy of nail lesion showed thickened nail plate, acanthotic epidermis, and atypical keratinocytes spanning the entire epidermis suggestive of BD of the nail [Figure 2c and d].

He was started on 5-fluorouracil 5% to be applied once daily at night for BD of genitals and podophyllin application once per week for verruca vulgaris. He showed remarkable improvement in both the lesions and there is no recurrence of nail lesions after 9 months of excision.

Discussion

BD is an intraepidermal SCC *in situ* predominantly occurring on sun-exposed areas; the most common sites being the head and neck, followed by lower limbs but it also affects photoprotected areas such as anogenital regions.[3]

The pigmented variant of BD is unusual. Pigmentation may be caused by human papillomavirus (HPV) of specific types which induces melanogenesis by releasing specific cytokines from neoplastic cells.[4,5]

It is a premalignant condition and the risk of progression to invasive carcinoma in extragenital lesions is 3%–5%, whereas in genital lesions it is 10%.[6]

Predisposing factors such as arsenic exposure, chronic sun exposure, HPV infection, and immunosuppression are commonly implicated in the pathogenesis of BD.[7]

There is a significant risk of SCC *in situ* in immunocompromised patients. In these patients, SCC *in situ* is likely to be multiple and more aggressive.[8]

BD of the nail unit is rare which can present clinically as verrucous lesion, fissuring, ulceration, onychocryptosis, paronychia, or nail dystrophy. Another rare presentation of the nail unit BD is longitudinal melanonychia which was first described by Baran and Simon in 1988. Fingernails are more commonly affected.[9,10] Although BD of the nail unit has a good prognosis, the incidence of local bone invasion in cases of invasive carcinoma of the nail unit ranges from 18% to 60%. The preferred therapeutic option is surgical removal with Mohs micrographic surgery being the treatment of choice.[11,12]

Association of HPV with the type of BD has been reported. HPV-associated BD mostly involves the anogenital region but approximately 30% of cases of HPV-associated extragenital BD have also been identified, especially caused by HPV type 16. The most common site of HPV-associated extragenital BD is the hand as the finger and nail unit are the most vulnerable sites for trauma. The anogenital-digital spread theory which states that viral transmission from the anogenital area to the periungual area is more reliable than that in the opposite direction. The reasons are as follows: first, there are higher infection rates of HPVs in anogenital cancer than that of periungual BD; second, the time sequence between the outbreak of vulvar lesions and changes of periungual lesions; third, the theory of autoinoculation from the anogenital area to the finger by scratching behavior. This theory best explains the co-occurrence of BD of genitals and nail unit as seen in our case.[2]

Our patient (immunocompromised) had coexistence of BD at two anatomically unrelated sites (genitals and nail unit) developed in succession. BD of the nail unit itself is rare and our patient had the rarest presentation as longitudinal melanonychia. BD of genital was of pigmented variety which is also uncommon. He also had verruca vulgaris of the penis suggesting that the cause of BD was HPV infection.

We are reporting this case, to the best of our knowledge, to date such a case has not been reported in the literature.

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Conflicts of interest

There are no conflicts of interest.

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