

## Extra-Genital Bowen's Disease on Abdomen Co-existing with Vulvar Intraepithelial Neoplasia

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### About the Author



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### Introduction

Bowen's disease was first described by the American dermatologist Dr. John T Bowen in 1912 as a form of intra-epidermal squamous cell carcinoma in situ of the genital region [1]. However, the International Society for Study of Vulvar Disease (ISSVD) in the latest classification of the neoplastic conditions of vulva has included Bowen's disease in the group of vulvar premalignant lesions termed as vulvar intraepithelial neoplasia (VIN). Lesions which have gross and histopathological appearance similar to Bowen's disease have been reported infrequently in the non-genital areas. This is known as extra-genital Bowen's disease (EGBD). Such lesions have been described in the exposed parts of the

body, specially in the lower part of leg and the periungual regions. Although majority of the VIN lesions are associated with oncogenic Human Papillomavirus (HPV) infection, the reported association between EGBD and HPV is variable [2]. In this article we report a case of HPV negative EGBD in an unusual site of the body along with VIN.

## Case Report

A 60-year old multiparous postmenopausal lady attended the Department of Gynecologic Oncology, Chittaranjan National Cancer Institute (CNCI) in January 2012 with chief complaints of pruritus vulvae and a gradually enlarging red patch on the perineal region for the last 2 years. She also complained of a slowly enlarging red patch on the skin of the lower abdomen. Nothing significant was found on general physical examination. On loco-regional examination, there was a pigmented, thickened lesion over the skin covering the labia majora and the labia minora, sparing the urethra, with signs of excoriation (Fig. 1). The lesion was extending up to inner aspect of thigh and perianal region. There were multiple wart-like projections all over the lesion. There was no ulcer or growth over vulva. Inguinal lymph nodes were not palpable. There was another distinct 4 cm × 3 cm lesion on the skin overlying the left iliac fossa (Fig. 2). This lesion was scaly, crusted, discrete, erythematous plaque with irregular border, and central atrophy. The lesion was not indurated. No mucocutaneous lesions were found anywhere else in the body.

Colposcopic examination of vulva revealed multiple dense acetowhite areas covering labia majora and minora extending to the fourchette. No abnormal vessels were seen on the lesion. Cervix was normal on colposcopy. All routine blood investigations were normal. Serology test for HIV1 and 2 and VDRL test were negative. Multiple punch biopsies obtained from the vulvar lesion confirmed VIN.



**Fig. 1** VIN affecting labia majora, minora, and adjacent skin



**Fig. 2** Extra-genital Bowen's disease on the skin overlying the left iliac fossa

The patient underwent simple vulvectomy along with wide local excision of the abdominal wall skin lesion with removal of 1 cm clinically healthy margin.

The histopathological examination of the post-operative specimen of vulva showed koilocytic changes in the superficial epidermal layer with squamous atypia in mid epidermis and basal layer with high mitotic count. The histopathological diagnosis was VIN (usual warty type) according to the ISSVD 2004 classification. Histological examination of the abdominal wall lesion showed hyperkeratosis and parakeratosis with koilocytic changes on the upper epidermis. There was full thickness epidermal atypia with mitosis of at least 10 cells per 10 high power fields in the basal and the mid-epidermal layers of the skin lesion. The histopathology diagnosis of the abdominal wall lesion was extra-genital focus of Bowen disease. There was no evidence of invasion, and the excised margins were clear of disease for both genital and extra-genital lesions.

The tissues extracted from the histology paraffin blocks of both vulvar and the extra-genital lesions were subjected to polymerase chain reaction (PCR) to detect HPV DNA using GP5 +/6 + primer for L1 consensus region of HPV. The PCR results were negative for HPV. The patient is on regular follow-up for the last 2 years and was disease-free at last contact.

An informed written consent was obtained from the patient to utilize her clinical materials for the present case report.

## Discussion

In the ISSVD 2004, nomenclature vulvar intraepithelial neoplasia (VIN) has been classified into “VIN usual type” and “VIN differentiated type.” A particular variant of

either of these conditions was earlier known as Bowen's disease.

However, this terminology is no longer used to describe vulvar pre-malignant lesions. Extra-genital Bowen's disease (EGBD) is still a rare but recognized cutaneous disease. EGBD is commonly associated with penile or vulvar pre-malignant lesions and affects the exposed parts of the body, mostly fingers, and the lower parts of the legs due to autoinoculation. The case reported by us is unusual in its site of affection. We observed the EGBD lesion in the lower abdomen, which is a non-exposed part of the body. EGBD has been reported in the non-exposed skin in cases of chronic arsenic exposure [3]. Our patient was a resident of Nepal, where till date, there has been no report of high arsenic content in drinking water. The patient did not have any other manifestations of arsenic induced toxicities.

Genital pre-invasive and invasive diseases have strong association with the oncogenic HPV infections. A meta-analysis observed that HPV is detected in 85.3 % of high-grade VIN lesions and in 40.4 % of vulvar carcinomas, with HPV 16 being the most common type [4]. Some of the studies demonstrated the presence of oncogenic HPV in EGBD lesions also [2]. However, the spectrum of HPV presentation and its detection in EGBD is still uncertain. In our case, no HPV was detected either in the VIN or in the EGBD lesion. Majority of the HPV positive VIN lesions are seen in women in their third or fourth decade of life, whereas our patient was much older, which may account for the HPV negativity.

The management of EGBD is mainly surgical along with appropriate management of the genital disease if any. We also observed good prognosis after surgical management.

## Conclusion

HPV negative EGBD can be seen in areas other than the extremities and even in non-exposed areas. The condition has good prognosis after surgery.

**Conflict of interests** None of the authors have any conflict of interest to declare.

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