Pyramid Journal of Medicine
Pyramid Journal of Medicine 2026; 9:540
*doi:10.4081/pjm.2026.*540

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Buschke-Löwenstein tumor of the vulva: case report and review of literature

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Abstract

Human papillomavirus is a common sexually transmissible infection, often asymptomatic. Where it does present with symptoms, they range from self-limiting anogenital warts (in low-risk subtypes) to malignant transformation (in high-risk subtypes).
In extremely rare circumstances, anogenital warts can progress to become giant condyloma acuminata, a condition known as Buschke-Löwenstein Tumor (BLT). BLT is a slow-growing locally aggressive, cauliflower-like benign tumor, with a tendency to malignant transformation. In some instances, it could co-exist with atypical epithelial cells or a well-differentiated squamous cell carcinoma. This tumor is usually resistant to conventional medical therapy for anogenital warts. Surgical resection is often the treatment modality, the extent of which depends on the extent of the lesion. We report a case of giant vulvar warts in a 17-year-old girl, who was successfully treated with skinning vulvectomy.

Key words: Buschke-Löwenstein tumor, human papilloma virus, giant condyloma acuminata.

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Introduction

Human papillomavirus is a common sexually transmissible infection, often asymptomatic, with a prevalence of up to about 32% in Nigerian women.[1] While the high-risk subtypes (16, 18, 31, 33 *etc*.) are associated with malignant transformation,[2] the low-risk subtypes (6-11) are usually associated with self-limiting anogenital warts (condyloma acuminata).[3] In extremely rare circumstances, in association with immunosuppression, these warts can progress to become Giant Condyloma Acuminata (GCA), a condition known as Buschke-Löwenstein tumor (BLT).[4] BLT is a slow-growing growing locally aggressive, cauliflower-like benign tumor, with a tendency to malignant transformation. In some instances, it could co-exist with atypical epithelial cells or a well-differentiated squamous cell carcinoma. This tumor is usually resistant to conventional medical therapy for anogenital warts. Surgical resection is often the treatment modality, the extent of which depends on the extent of the lesion.[4] We report a case of giant vulvar warts in a 17-year-old girl, who was successfully treated with skinning vulvectomy.

Case Report

A 17-year-old girl presented to our gynaecological outpatient department on 30/11/2021 with a 3-month history of a progressive, painful growth on her genital area. It was insidious in onset, initially limited to the left labium majus, which later progressed to cover the whole perineum, large enough to prevent her from sitting or walking properly. There was associated offensive discharge.

She was sexually active, both heterosexual and homosexual. She was not a known diabetic, not on any immunosuppressive medication and not a known retroviral disease patient. She had not sought care earlier as she was afraid to inform her parents.

On examination, she was anxious and in painful distress. Genital examination revealed a giant dark greyish-pinkish cauliflower-like mass with fungating surfaces composed of multiple coalescing smaller masses, involving the whole of the left labium majus, fourchette, right labium majus and mons pubis measuring 15 by 13 cm. Tenderness precluded deeper examination (Figure 1). There was no peripheral lymphadenopathy.

A small sample was excised for biopsy. Retroviral, diabetes, and hepatitis screening were all negative. She was placed on antibiotics, sitz baths and topical podophylin. A week later, she returned with a less fungating mass, but there was no regression in size. The histology report revealed condylomata acuminata.

She was then booked for examination under anaesthesia and definitive surgery (20/12/2021). Intraoperatively, a more extensive examination of the mass revealed isolated smaller lesions on both labia minora, the tip of the clitoris and lower third of the posterior vaginal wall. The urethral orifice, perianal region and anal orifice were not involved.

She had a skinning vulvectomy done successfully (Figure 2). The smaller lesions on the clitoris and labia minora were cauterized. Two units of blood were transfused intraoperatively.

The postoperative course was successful. She however, devel-

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oped a surgical site infection and wound breakdown around the reconstructed fourchette on the 7th postoperative day, which was allowed to heal by secondary intention after sitz baths and oral antibiotic administration.

Histology of the excised mass (Figure 3) revealed: a huge polypoid skin mass measuring 13 cm across and weighing 180 g. Microscopy revealed a skin lesion composed of epidermis disposed in papillary patterns and lined by epithelial cells exhibiting koilocytosis, acanthosis, hyperkeratosis and parakeratosis, overlying an unremarkable fibromuscular core.

She was followed up at 2 weeks, 6 weeks (Figure 4) and 3 months postoperative. The lesion healed with a posterior perineal defect measuring 3 cm. She was counseled on ways of preventing sexually transmissible infections. She is currently scheduled for a yearly follow up.

She had a colposcopy done 10 months later to rule out coexisting malignancy, which was normal. She has remained recurrence-free up to 3 years post-operative.

Discussion

Only 10% of women infected with Human Papillomavirus (HPV) develop anogenital warts.[5] In extremely rare circumstances, anogenital warts may progress to GCA (BLT). This occurs most often in men and in immunocompromised states like Human Immunodeficiency Virus (HIV).[4] Our patient was screened for HIV and diabetes but turned out negative; however, her low socioeconomic status could have contributed to the impaired immune response.

GCA is a slow-growing, locally destructive verrucous lesion that typically appears on the penis but may occur elsewhere in the anogenital region. Few cases of vulvar GCA have been reported.

It most commonly is considered to be a regional variant of verrucous carcinoma.[6] It could coexist with an atypical lesion,[7] well differentiated squamous tumor or later progress to a squamous tumor. However, some authors do consider it and verrucous carcinoma to be separate entities: GCA with no malignant potential and the other with the ability to metastasize.[6]

In this patient there was no co-exisiting tumor and follow up colposcopy showed normal findings.

Histologically, it shows pseudo-epitheliomatous proliferation and local invasion by massive epidermal hyperplasia, hyperkeratosis, and parakeratosis; It is also markedly exophytic.[8]

Although the exact pathogenesis is not clear, the E6 protein of HPV-6 and HPV-11 binds p53 tumor suppressor protein less effi-

Figure 1. Pre-operative.

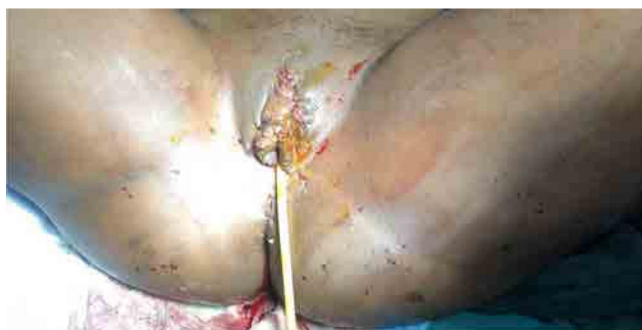

Figure 2. Immediate post-operative.

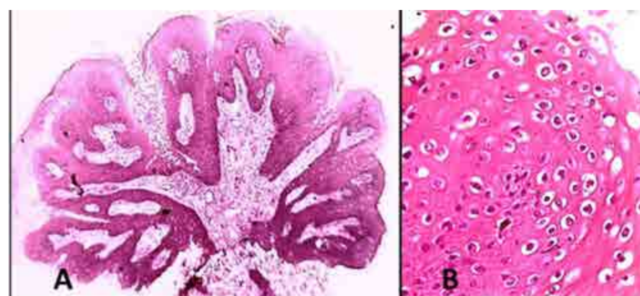

Figure 3. Mass on histology.

Figure 4. Six weeks post-operative.

ciently than that of HPV-16 and HPV-18 but, theoretically, could lead to rapid degradation of the p53 protein. The E6 protein also inhibits p53 transcription.

A mutation in p53 protein leading to clonal proliferation has been suggested. Other implicated agents are chronic chemical exposure, chronic irritation, and poor hygiene.[5-7]

The growth starts as a keratotic plaque that expands slowly into a cauliflower-like mass. It takes longer in immunocompetent individuals. In immunosuppressed individuals, however, the growth is commonly more rapid.[9] The lesion may then ulcerate; typically, the lesion may be associated with foul odor like in our case. Regional lymph node enlargement is common usually following secondary infection; our patient, however, did not have enlarged lymph nodes.[5-7]

Due to the rarity of this tumor, there is no standard treatment. Wide surgical excision has been considered treatment of choice to prevent recurrence.[10-12] Surgical excision alone has been shown to result into disease-free state in most cases.[12] Oral and topical chemotherapeutic modalities have been used alone or as adjuvants, to surgery.[13]

Conclusions

BLT is a rare vulvar tumor that can be treated successfully with wide local excision. However, due to its relationship with verrucous carcinoma of the vulva, a coexisting tumor has to be ruled out and patients must be followed up postoperatively to ensure they remain recurrence-free.

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Received: 28 July 2025; Accepted: 23 September 2025.

Contributions: HDA conceived and designed the work, reviewed it critically for important intellectual content; AAB co designed the work, drafted the manuscript. Both authors have read and approved the final version of the manuscript, and agreed to be held accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Conflict of interest: the authors declare no potential conflict of interest.

Funding: none.

Ethics approval and consent to participate: not applicable.

Informed consent: not applicable.

Patient's consent for publication: the patient gave her written consent to use her personal data for the publication of this case report and any accompanying images.

Availability of data and materials: all data generated or analyzed during this study are included in this published article.

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