International Journal of Medical Science and Clinical Research Studies

ISSN(print): 2767-8326, ISSN(online): 2767-8342

Volume 04 Issue 08 August 2024

Page No: 1478-1483

DOI: https://doi.org/10.47191/ijmscrs/v4-i08-11, Impact Factor: 7.949

Buschke-Löwenstein Tumor in HIV-Positive Patient, Surgically Treated by V-Y Advancement Flap, Case Report and Review of the Literature

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ABSTRACT

Introduction: Buschke-Löwenstein tumor (BLT), known as giant condyloma acuminatum, is a sexually transmitted disease associated with human papillomavirus (HPV), mainly genotypes 6 and 11. The frequency of occurrence is rare and it is mainly located in the external genitalia and perineal region.

Among the risk factors associated with this tumor are immunosuppression, alcoholism, smoking, the presence of genital infections, among others. Histologically, the distinctive feature of BLT is the lack of involvement of the basement membrane; however, it is said that this tumor presents mixed characteristics of squamous cell carcinoma and condyloma acuminatum.

Clinically it manifests as a slow growing tumor, pain, itching and tends to present ulcerations, fistulas or bleeding as well as high risk of recurrence. Surgical resection with free margins and reconstruction is the gold standard of treatment, however there are multiple options and treatment must be individualized, taking into account the risk of recurrence and cancerous progression to squamous cell carcinoma.

Case presentation: We present the case of a 24 year old male patient, HIV positive, with a giant condyloma acuminatum in the perianal region, affecting the anal canal and gluteal folds. Surgical resection of the tumor was performed with bilateral V-Y advancement flap reconstruction in both gluteal folds. The postoperative period was managed without serious complications and the patient showed a favorable evolution.

Conclusion: Although surgical resection with reconstruction is the gold standard for the treatment of Buschke-Löwenstein tumor, treatment must be individualized due to the rarity of this pathology, especially in immunocompromised patients. In our case, the patient was treated by resection and V-Y advancement flap, achieving recovery without major complications.

ARTICLE DETAILS

Published On:
10 August 2024

Available on:
https://ijmscr.org/

INTRODUCTION

Buschke-Lowenstein tumor (BLT), also known as giant condyloma acuminatum, is a sexually transmitted disease, associated with human papillomavirus infection, mainly genotypes 6 and 11. Abraham Buschke first reported it in 1896. Later in 1925 Abraham Buschke together with Ludwig Löwenstein described it as a lesion with mixed features of squamous cell carcinoma and condyloma acuminatum, but with different biological and histopathological characteristics.1,2

Risk factors associated with BLT include smoking, alcoholism, chronic local inflammation, immunodeficiency (HIV-diagnosed individuals, transplant recipients and diabetics), pregnancy, chronic genital infections, multiple sexual partners and suboptimal hygiene practices. 2,3,4

BLT is a well-defined tumor with basement membrane involvement being the main difference between a Buschke-Löwenstein tumor and a squamous cell carcinoma. Histologically BLT is characterized by significant epithelial hyperplasia, sometimes pseudoepithelioma with intact basement membrane, hyperacanthosis, hyperpapillomatosis and koilocytosis. 2,5

Buschke-Löwenstein tumor is located mainly in the external genitalia, in men the most frequent location is in the penis and in women in the vulva in 90% of cases, it can also occur in the anorectal region and rarely in the bladder and axilla. It manifests clinically as a slow-growing tumor, pain, itching, constipation and difficulty urinating. Lesions tend to form fistulas, ulcers, bleeding, abscesses and recurrence; rarely, inguinal lymphadenopathy occurs, which is associated with inflammation and infection caused by the tumor.6,7,8

Diagnosis is achieved by performing a detailed clinical history and examination, however for more developed cases preoperative imaging studies may be required to delimit the depth and extent of infiltration and to plan surgical treatment. Histological confirmation should also be obtained by taking an adequate biopsy which should be wide and deep enough to obtain accurate images to detect basement membrane infiltration, lymphovascular invasion and distant metastasis.9

Treatment of BLT encompasses various options, such as podophyllin, 5-fluorouracil, methotrexate, bleomycin, cryotherapy, interferon, curettage, CO2 laser vaporization, chemotherapy, radiotherapy and wide surgical resection. Surgical resection with free margins and reconstruction remains the gold standard, however due to the rarity of the pathology a standardized approach to choose the therapeutic approach has not yet been established, recommending an individualized approach for each case.9,10,11

During surgical resection, free margins of at least 1 cm should be ensured and subcutaneous fatty tissues should be included. In those cases located in the perineum, one should always try to preserve and reconstruct the anal sphincter. Regarding reconstruction techniques, VY fasciocutaneous advancement flaps, S-flaps or island flaps can be used, being especially difficult in the perineal region, because they can present more complications such as additional wounds, bleeding, hematoma, wound infection or dehiscence.7,10,12

Another radical surgical procedure that can be performed is abdominoperineal resection with terminal colostomy which is chosen in those patients in whom the anal sphincter is invaded and they present incontinence, diffuse invasion of the rectal and lateral pelvic wall, who present multiple recurrences and malignancy. In cases where TBL transforms into squamous cell carcinoma, surgical resection can be combined with radiotherapy and chemotherapy as neoadjuvant or adjuvant therapy.10,13

Recurrences are a common feature of Buschke-Löwenstein tumor especially after incomplete resection and due to the risk of malignant transformation to squamous cell carcinoma ranging from 30% to 56% it is important to follow up closely after initial treatment. 13,14,15

EPIDEMIOLOGY

Buschke-Löwenstein tumor is a rare disease although its precise incidence is unknown; it is estimated to affect approximately 0.1% of the general population, it can present at any age, especially between 30 and 50 years of age with a mean age of onset of 40 years; several cases have been reported in pediatric age, and sexual abuse should be suspected in these case. 1, 16-18

Buschke-Löwenstein condyloma affects men more frequently than women, with a 3:1 ratio; a higher incidence has been observed in patients with immunodeficiency, homosexuals and bisexuals. With respect to mortality, rates of 20-30% have been reported, however these figures are in doubt since these deaths were related to infectious complications.19,20, 21

PHYSIOPATHOLOGY

Buschke-Löwenstein tumor is a sexually transmitted disease that can also be transmitted horizontally and vertically. It is mainly caused by HPV infection, especially by subtypes 6 and 11, which contribute to more than 90%, and exceptionally by subtypes 16 and 18. Initially the disease presents from a condyloma acuminatum of long evolution, whose diameter can exceed 10 cm. The time from the appearance of the first symptoms to the development of BLT ranges from 2.8 to 9.6 years. 1,18

Papillomaviruses are DNA viruses of the Papillomaviridae family, with tropism for stratified squamous epithelia. HPVs are classified into α, β, γ, μ, and ν; α-HPVs are subdivided according to their oncogenic potential into low-risk (HPV-6, HPV-11) and high-risk (HPV-16, -18, -31, -33, -35, -39, -45, -51, -52, -56, -58, -59, -66, -68, -70). Low-risk types are associated with condylomas while chronic infection with high-risk types is the main factor for HPV-induced malignant neoplasms, especially HPV-16, which causes the majority of anogenital squamous cell carcinomas. 22,23,24

Once HPV infects basal keratinocytes, the viral DNA remains in episomal form. Although the immune system usually eliminates the infected cells, in some cases, the infection becomes chronic. HPV replication in epithelial cells increases the expression of E6 and E7 oncoproteins, which induce genomic instability, cell cycle alteration, proliferation, immortalization and malignant transformation as well as immunosuppression. 15

Cell-mediated immunity is essential to control HPV-induced lesions therefore patients with immunosuppression or HIV are at increased risk of severe HPV infections as well as high risk of progression to metastatic squamous cell carcinoma. CD4+ T cells and monocytes/macrophages predominate within regressing condylomas. Akinboro et al. documented lower blood CD4+ cell counts in HIV-positive patients with genital warts compared to those without lesions and also

found correlations between the extent of warts and CD4+ T cell counts. 25

CASE PRESENTATION

Male patient 24 years old, HIV positive for 1 year in control with Bictegravir / emtricitabine / tenofovir alafenamide once a day, refers to have anoreceptive sexual intercourse, protecting himself with condoms, the rest of the history denies them.

His condition began 1 year ago with the presence of a wart-like lesion in the perianal region, asymptomatic with intermittent evolution, managed with home remedies without resolution. He went for medical evaluation and was diagnosed with HIV with decreased cd4+ load. The patient presented worsening of the lesions with the appearance of abundant condyloma acuminata in the anal and perineal region and rectorrhagia on defecation.

During the coloproctology evaluation, giant condyloma acuminatum was found in the perianal region with extension of the lesions to the anal canal and gluteal folds, so it was protocolized for surgical resection due to the suspected diagnosis of buschke-lowenstein tumor. Pre-surgical studies were taken: WBCs 6.10 N 64.3%, CD4 lymphocyte count: 470 cells per cubic millimeter Hemoglobin 12.9 g/dL, platelets 331 thousand, HIV viral load was undetectable.

Surgical resection of the tumor and reconstruction by anoplasty with V-Y flap advancement was performed, during the surgery giant condylomas were found in perianal region with circumferential extension to the anal canal and extension to the right and left gluteal fold. The anal sphincter was respected and the anoplasty was performed circumferentially with advancement of the V-Y flap, the drainage was closed bilaterally in both buttocks.

In the postoperative period the patient was managed with closed drainage, analgesia, antiretroviral treatment, antibiotic therapy with double scheme, without complications, he was discharged home after removal of the drainage, during the follow-up he presented scarce seropurulent material in the surgical wound, which merited antibiotic therapy, cures with adequate evolution, the result of the biopsy was collected reporting compatible findings of condyloma acuminatum without data of malignancy so it only continued in follow-up and antiretroviral treatment.

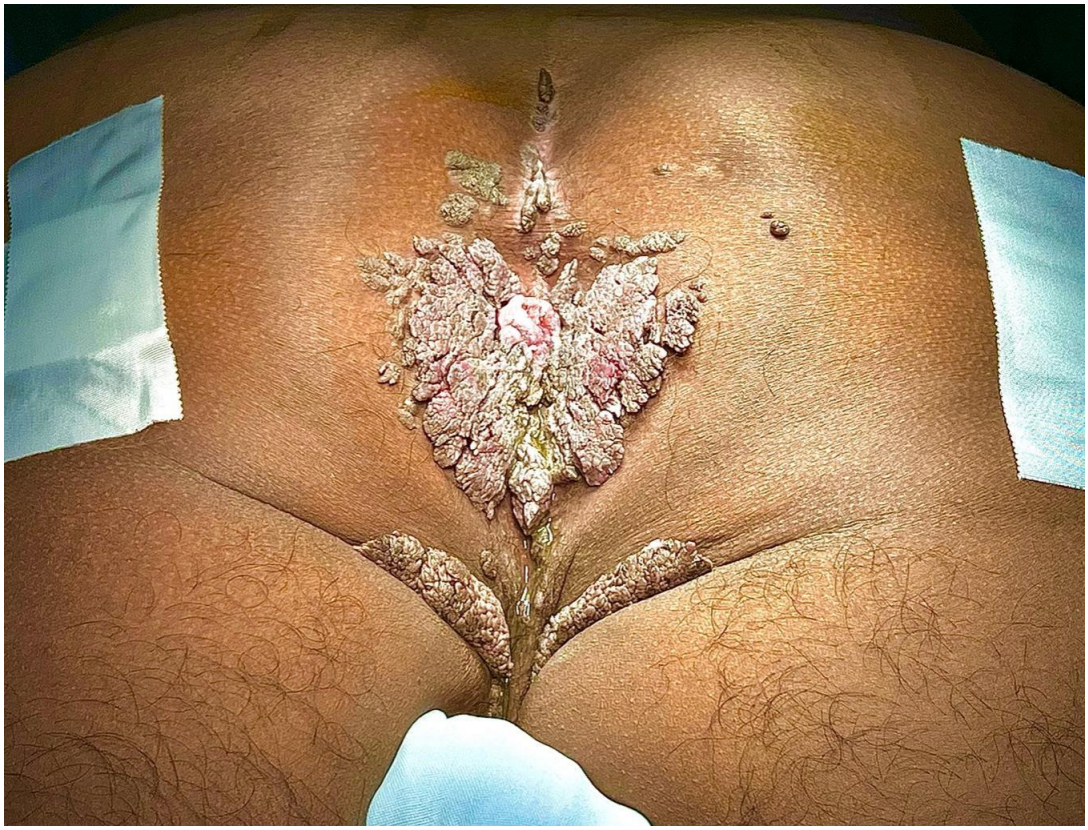

Figure 1. Buschke-Löwenstein tumor in the perianal region invading the anal canal and gluteal folds.

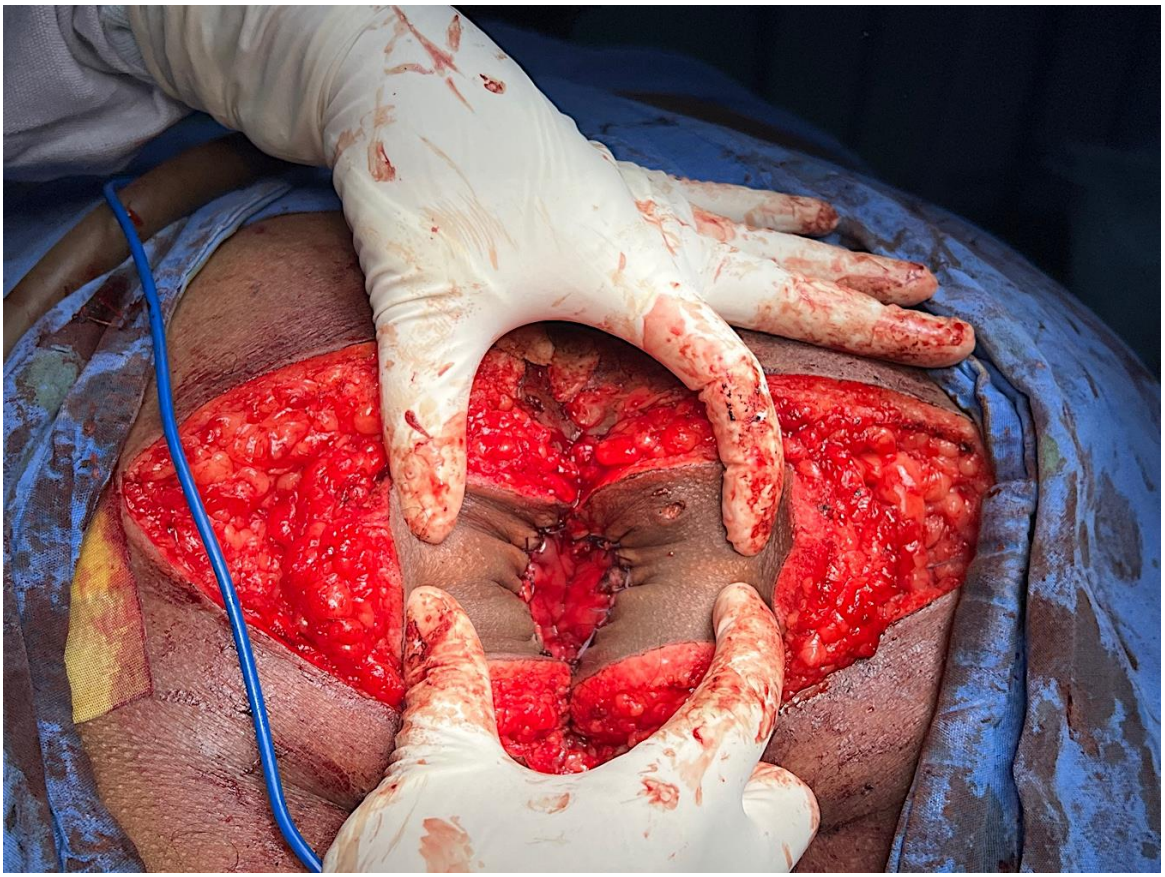

Figure 2. Intraoperative photograph after wide resection and circumferential anoplasty with VY flap.

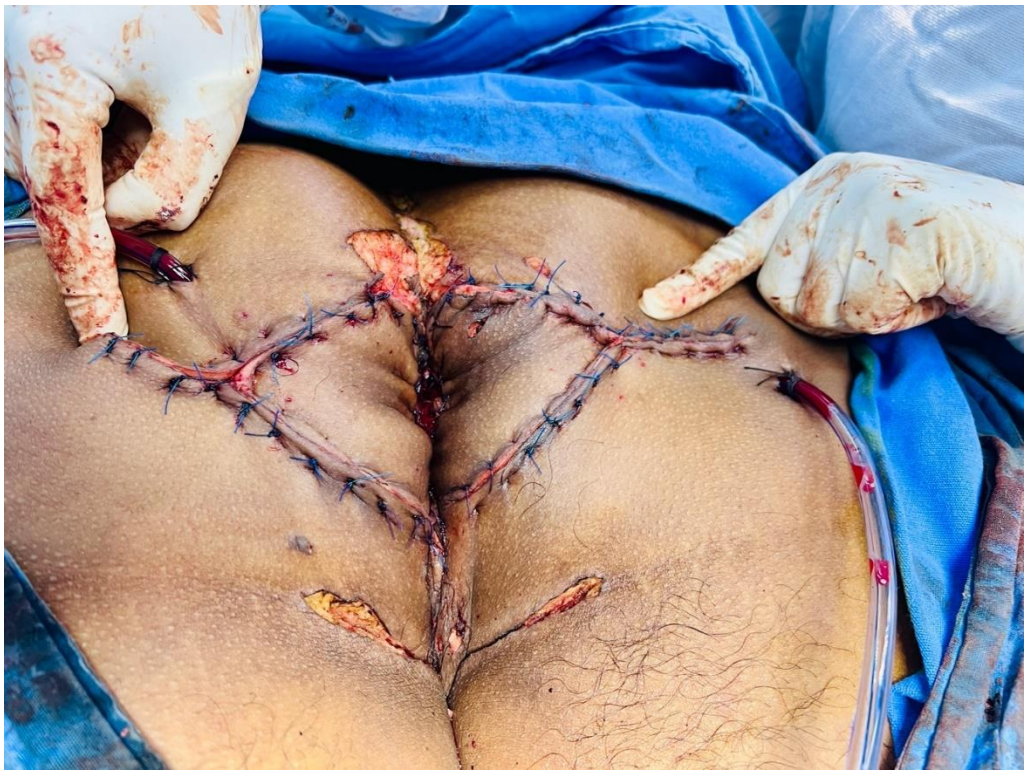

Figure 3. Immediate postoperative picture of bilateral V-Y flap with closed drain placement in both buttocks.

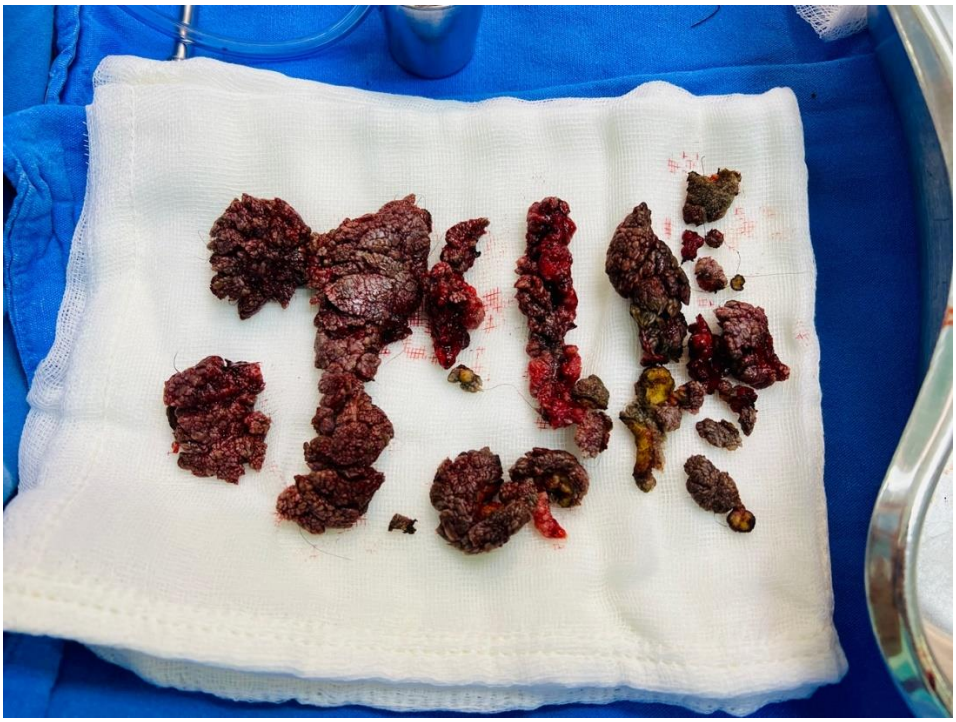

Figure 4. Surgical specimen after removal.

CONCLUSIONS

The management of Buschke-Löwenstein tumor (BLT), a rare and locally aggressive variant of anogenital condylomata acuminata, presents significant challenges in clinical practice. This condition, caused by persistent infection with human papillomavirus (HPV), predominantly types 6 and 11, exhibits a propensity for extensive local invasion and recurrence, necessitating a multidisciplinary approach for optimal patient outcomes.

Surgical intervention remains the cornerstone of treatment for BLT. Wide local excision, often coupled with reconstructive procedures, is critical to achieving clear margins and minimizing the risk of recurrence. The complexity of surgical management is heightened by the tumor's propensity for infiltrative growth and the anatomic intricacies of the affected regions, which frequently involve the anogenital and perineal areas. Despite the efficacy of surgical excision, the recurrence rate remains substantial, necessitating vigilant postoperative surveillance.

Adjunctive therapies, including laser ablation, cryotherapy, and topical agents, have been employed with varying degrees of success. These modalities are particularly useful in cases where surgery is contraindicated or when residual disease is present postoperatively. The utilization of carbon dioxide (CO_2) laser, in particular, offers a precision modality that minimizes damage to surrounding healthy tissue, although it requires careful patient selection and expertise.

Recent advancements in systemic therapies, such as the use of interferon and imiquimod, have shown promise in treating BLT. These agents, which modulate the immune response, may reduce the viral load and induce regression of lesions. However, their role remains adjunctive, and further studies are needed to delineate their efficacy and safety profile in conjunction with surgical treatment.

The advent of HPV vaccination has introduced a potential preventive strategy for BLT. Vaccination against HPV types 6 and 11 has been shown to reduce the incidence of genital warts and, by extension, may decrease the occurrence of BLT. This preventive measure underscores the importance of public health initiatives aimed at increasing vaccination coverage, particularly in high-risk populations.

In conclusion, the management of Buschke-Löwenstein tumor requires a comprehensive, multidisciplinary approach that integrates surgical, adjunctive, and systemic therapies. The aggressive nature and high recurrence rate of this tumor necessitate ongoing research into innovative treatment modalities and preventive strategies. Moreover, patient education and regular follow-up are essential components of care to ensure early detection of recurrences and to address the psychosocial impacts of this condition. As our understanding of HPV-related diseases continues to evolve, the future of BLT management holds promise for improved therapeutic outcomes and enhanced quality of life for affected individuals.

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