Case Report

Bushke-Lowenstein Tumor, First Giant Tumor Reported in Ecuador

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Abstract

Describe a clinical case of Buschke-Löwenstein Tumor and its surgical management in a tertiary hospital.

Introduction

Buschke-Löwenstein tumor (BLT), also known as giant condyloma acuminata, was first described in 1925 by Abraham Buschke and Ludwig Löwenstein as a neoplasm of the penis that resembled both a condyloma acuminata and a squamous cell carcinoma [1]. BLT is a rare exophytic tumor that primarily affects the anogenital and perianal regions. It is strongly associated with low-risk human papillomavirus infection [2]. It is characterized by its locally aggressive behavior, potential for destructive growth, and high recurrence rates, despite its benignity [3]. BLT is a rare pathology and there is no clear therapeutic guideline, although wide surgical excision with clean margins is considered primary treatment to minimize the risk of recurrence [4,5]. Various complementary therapies have been explored, including laser, cryotherapy, radiation therapy, electrocoagulation, and immunotherapy, but surgery remains the cornerstone [4]. In cases where surgery is not feasible or in patients with significant comorbidities, alternative treatments are considered [6].

Materials and Methods

We present the case of a 59-year-old male patient, hospitalized in the Urology service of the Eugenio Espejo Specialty Hospital in the city of Quito, Ecuador, in the month of June 2021 due to an exophytic, condylomatous lesion located in the genital area, pubic, perianal and inner thighs bilaterally, approximately 25 cm long x 12 cm wide. According to the patient's verbal references, this lesion progressively increased in size for almost 2 years. The perianal mass was completely removed, ensuring negative surgical margins. The large perineal, scrotal, and perianal skin defect that occurred was reconstructed with multiple VY fasciocutaneous advancement flaps and skin grafts. The need for a protective stoma arose. The literature review extended from December 2006 to September 2023, using Pubmed and Google Scholar as search platforms.

Presentation of the Case

This is a 59-year-old male patient who came to the Emergency Department of the Eugenio Espejo Specialty Hospital with giant verrucous lesions of 2 years of evolution, located in the inguinal, perineal, gluteal, pubic and external genitalia regions, involving the scrotum and foreskin, without suppuration at the time of examination (Figure 1). These lesions were going to be treated at another hospital, but the procedure was postponed after an incidental finding in the presurgical blood count that revealed leukocytosis with evidence of lymphocytosis and monocytosis.

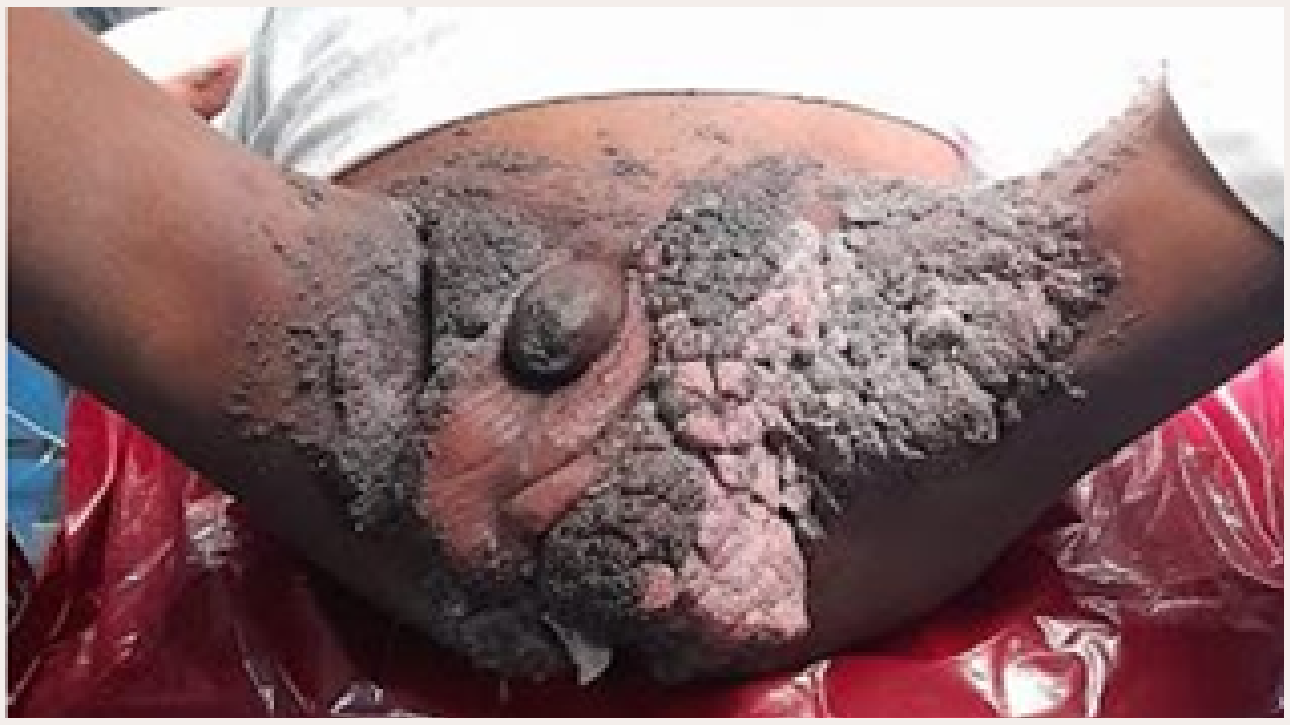

Figure 1: Anogenital and perianal regions before performing en bloc excision of the lesion.

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It was decided to hospitalize the patient for the study of the lymphoproliferative process and for the surgical treatment of warty lesions. After the study by the Hematology service, a diagnosis of chronic lymphoblastic leukemia was reached, who decided to defer the treatment of this pathology until treatment of the warty lesions. During hospitalization, the lesions began to emit a greenish, foul-smelling discharge. A tissue culture from the genital area was performed, which showed colonization by two complex bacteria, Klebsiella pneumoniae KPC ss. sensitive to Amikacin and Pseudomonas aeruginosa sensitive to Amikacin, Ciprofloxacin, Gentamicin, Imipenem and Meropenem, so Infectology assesses and decides on antibiotic therapy according to the results of the antibiogram.

The lesion was excised in its entirety, sending it for histopathological study, where tissue measuring 30 cm long by 23 cm wide was reported, with free lateral edges and free deep edges at 0.3 cm, respecting the mucosal foreskin. where no lesions were found, corresponding microscopically to a verrucous squamous cell carcinoma (Buschke-Löwenstein tumor), in addition, signs of HPV viral infection and a mild to moderate lympho-plasmacytic inflammatory infiltrate (Figure 2).

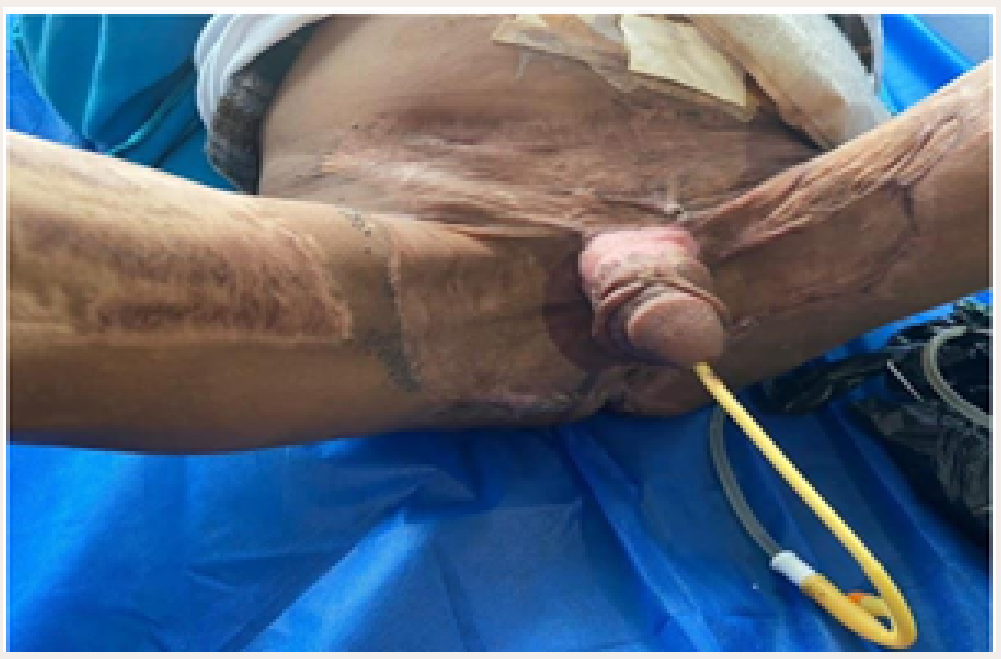

Figure 2: Appearance of the perineal and inguinoscrotal regions 6 months after treatment with placement of grafts taken from the thigh.

A multidisciplinary intervention was carried out on the case, the mass was completely removed between the Urology and Coloproctology services, by means of En bloc excision of the large diameter lesion and electrocoagulation of the peripheral lesions (Figure 3) plus construction of a protective colostomy. In a second stage, bilateral pockets were made on the antero-internal aspect of the thighs to cover the testicles and grafts were placed in the bloody area by the health service. Plastic Surgery, taken from the anterolateral aspect of the thighs, using dermatome (Figure 4).

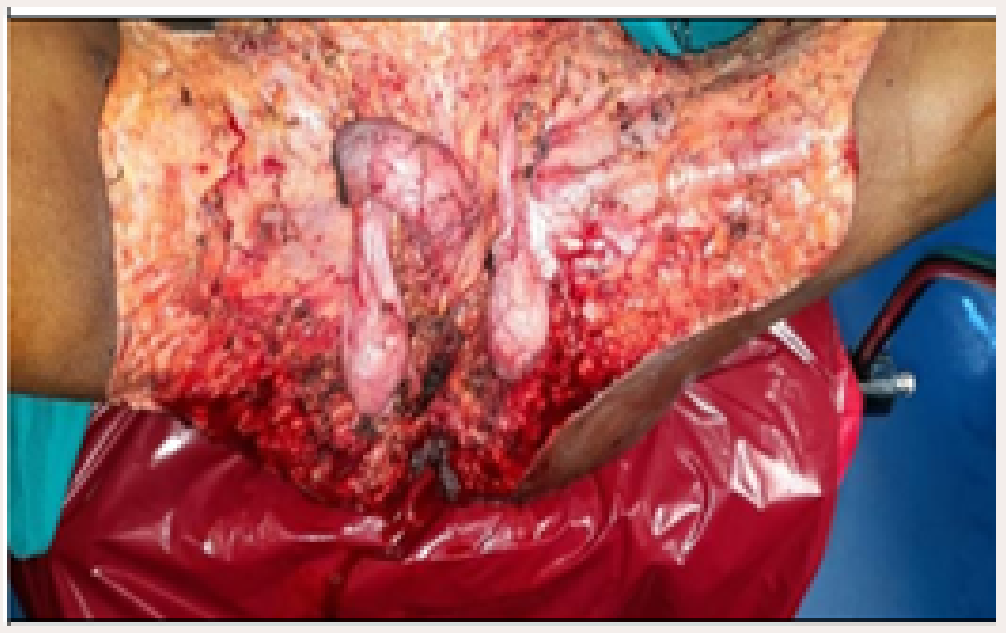

Figure 3: Appearance of the genital, perineal and inguinal regions after excision of a verrucous lesion.

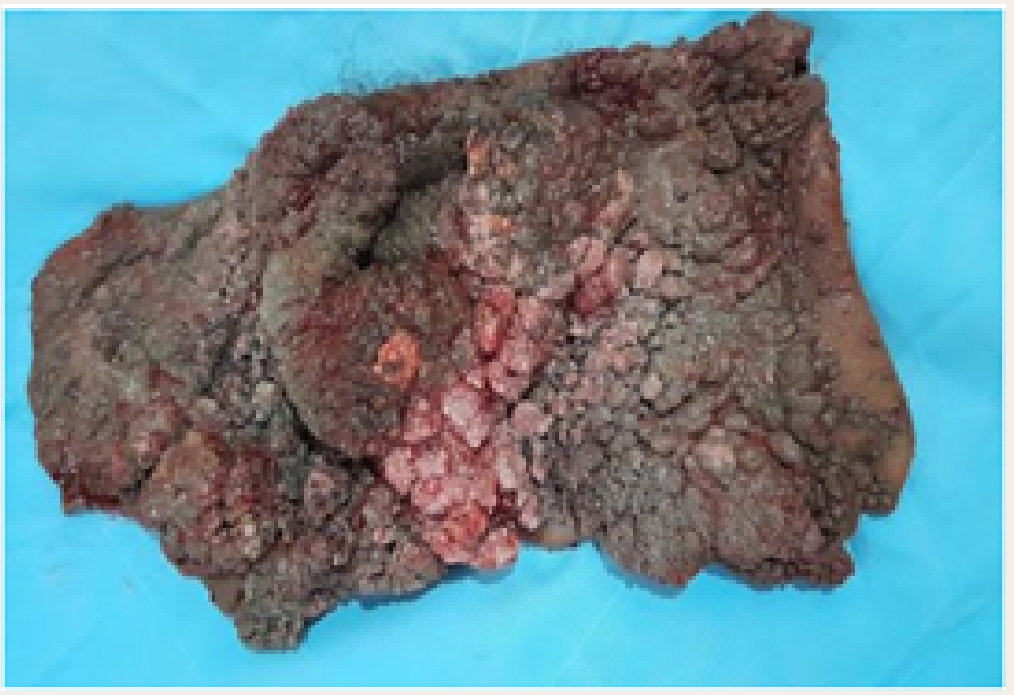

Figure 4: Piece surgically removed in block whose measurements were 30 cm long x 23 cm wide.

After treatment and discharge, the patient continued to undergo controls in Urology and Dermatology for electrofulguration of newly appearing lesions on the distal edges of the graft, given that this pathology has a high risk of recurrence. In addition, follow-up by oncohematology who initiated and maintains the patient in controls due to his diagnosis of Chronic Lymphoblastic Leukemia.

Discussion

Buschke-Lowenstein tumor is a rare entity. This incidence is not exactly known. However, it is believed to be higher than reported [7]. It was initially described as a tumor limited to the penis by Buschke in 1896 and Lowenstein in 1925. However, it can appear in multiple locations. The most common are the genital area and the perineal region, although other unusual areas such as the bladder have been described [8].

Clinically, BLT presents as giant, exophytic, warty lesions that destroy the tissues in which they reside, with a tendency to invade deeply. However, upon histological analysis it has a benign behavior, as it does not present signs of malignancy, such as infiltration or rupture of the basement membrane, lymphatic invasion, vascular invasion or distant metastatic potential. It is considered an intermediate entity between condyloma acuminatum and verrucous carcinoma, which explains its high morbidity and mortality. The average age of diagnosis is around 47.6 years, with a higher prevalence in men compared to women. Risk factors include smoking and immunosuppression, such as HIV infection or immunosuppressive therapy [9,10].

Both lesions may also coexist in up to 50% of cases. The rate of malignant transformation reaches 20% in the case of condylomata acuminate. However, for TBL it reaches up to 56% [11]. The relationship between condylomata acuminata and human papillomavirus (HPV) is well known, since this virus has been identified in more than 90% of condylomatous lesions, mainly in types 6 and 11. In contrast, types 16 and 18 are usually present in lesions with a high degree of dysplasia, carcinoma in situ and invasive carcinoma, indicating a higher risk of malignancy [4].

The histopathological study is necessary to make the diagnosis of the disease, since the microscopic examination reveals a leafy mass composed of wide and rounded epidermal ridges, where well-differentiated squamous cells are observed that do not show cellular anaplasia, surrounded by acute and inflammatory cells. chronicles [12,13].

This tumor has been treated in multiple ways, with radical cutting of the lesions and secondary healing or plastic reconstruction whenever feasible being the most appropriate surgical alternative [14]. Other methods have been used such as abdomino-perineal amputation (AAP) and lateral discharge colostomy. AAP is generally reserved for situations of tumor recurrence, pelvic invasion, or malignant change. In this clinical case, radical excision was chosen, since it is considered the technique that has the least recurrences. AAP was not necessary as there were no signs of malignancy in the pathological investigation nor in the recurrence. Colostomy is beneficial to prevent the danger of fecal contamination in the surgical lesion; However, it is not necessary in all cases [5,15-17].

Cryosurgery, intralesional Bleomycin and laser resection have been used, sometimes radiotherapy and systemic chemotherapy based on combinations with methotrexate, 5-fluorouracil, bleomycin, mitomycin C, cisplatin and leucovorin. However, there is no evidence in the literature that supports their systematic use. The use of intralesional or systemic interferon has also been reported [4].

The recurrence rate of BLT varies between studies. In a multicenter analysis of 38 cases, a recurrence rate of 23.7% was reported after a follow-up of 23 months [9]. Another study mentions a high recurrence rate after surgical excision, although it does not specify an exact percentage [4]. Furthermore, it has been reported that recurrence may be more frequent in cases where a complete resection is not achieved or in cases of chronic non-communicable diseases [18].

Conclusion

Buschke-Lowenstein tumor (TBL) is a rare pathology, secondary to a sexually transmitted disease caused by the human papillomavirus (HPV), described as an intermediate form between a condyloma acuminata and a squamous cell carcinoma.

It is found in the genital and anorectal area, has the capacity for malignant transformation and a high recurrence rate. Surgery is the first-line treatment, surgical treatment of Buschke-Löwenstein tumors in Ecuador requires a multidisciplinary approach, with a team of highly qualified professionals. The complexity of reconstruction techniques can carry additional risks, underscoring the importance of experience in this field. We believe that aggressive surgery, including reconstructive procedures, should be considered the standard treatment for these tumors. This approach seeks not only to achieve optimal surgical results, but also to prevent recurrence, thus contributing to a better quality of life.

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