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A Case of Buschke-Löwenstein Tumor: Diagnosis and Surgical Management Buschke-Löwenstein Tumor: A Case Report of Diagnosis, Surgical Treatment and Adjunctive Measures for Recurrence Prevention

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ABSTRACT

Buschke–Löwenstein tumor (BLT), also known as giant condyloma acuminatum or verrucous carcinoma, is a rare sexually transmitted disease associated with human papillomavirus (HPV) infection, predominantly HPV types 6 and 11, which account for approximately 90% of cases. It occurs mainly in males, with an estimated overall incidence of 0.1%. Despite its benign histological appearance, BLT is characterized by marked local aggressiveness, a capacity for extensive and destructive growth, a high recurrence rate following surgical excision and a significant risk of malignant transformation reported in up to 56% of cases.

We report the case of a 50-year-old patient with no significant medical history who presented with a large exophytic perineal lesion evolving over five years, accompanied by recurrent bleeding and pain. Clinical examination revealed multiple exophytic, vegetative, blackish tumors involving the penile shaft, glans and scrotum, measuring between 1 and 10 cm, with a characteristic cauliflower-like papillomatous appearance.

Histopathological examination demonstrated papillomatous epidermal hyperplasia with koilocytic vacuolization, confirming the diagnosis.

Radical surgical excision was performed, followed by four sessions of chemical destruction using 50% trichloroacetic acid and imiquimod, with close clinical follow-up. No recurrence was observed after one year,

emphasizing the importance of early diagnosis, complete surgical management and rigorous monitoring to optimize outcomes.

Multiple therapeutic modalities have been described in the literature, including laser therapy, cryotherapy, radiotherapy, electrocoagulation, immunotherapy, topical agents such as imiquimod and sinecatechins, intralesional 5-fluorouracil, isolated perfusion and systemic or local chemotherapy. In extensive cases, neoadjuvant chemotherapy or radiotherapy may be considered to reduce tumor volume and facilitate safer surgical debulking. HPV vaccination, which includes HPV types 6 and 11, significantly reduces the incidence of genital warts and may consequently decrease the risk of developing BLT, underscoring the critical role of HPV infection in the pathogenesis of this rare tumor.

Keywords: Buschke–Löwenstein tumor; Human Papillomavirus; Blackish Tumors

INTRODUCTION

Buschke–Löwenstein tumor (BLT), also referred to as giant condyloma acuminatum, is a rare sexually transmitted disease caused by human papillomavirus (HPV) infection [1], predominantly involving the external genitalia and/or the anorectal region. Although generally considered a benign lesion, BLT is characterized by locally aggressive behavior, a substantial risk of recurrence after treatment and the potential for malignant transformation [2]. Owing to the absence of standardized diagnostic criteria and the rarity of the condition, its exact incidence and prevalence remain difficult to define; however, the overall prevalence has been estimated at approximately 0.1% in the general population. BLT most commonly affects individuals in mid-adulthood, typically between the fourth and sixth decades of life, though sporadic cases have also been reported in pediatric patients and during pregnancy [3-6].

Epidemiological studies indicate a clear male predominance, with a reported male-to-female ratio of approximately 2.7:1 and a mean age at diagnosis of around 44 years. HPV plays a central role in the pathogenesis of BLT, with low-risk genotypes 6 and 11 accounting for more than 90% of cases in both immunocompetent individuals and those with underlying immunosuppression and, less frequently, high-risk genotypes 16 or 18, which are responsible for the notable tendency toward malignant transformation, typically resulting in squamous cell carcinoma (SCC) [7-9]. Identified risk factors include multiple sexual partners, chronic genital infections, inadequate hygiene, smoking and immunodeficiency states. While HPV vaccination has demonstrated a protective effect by reducing the incidence of genital warts, data specifically addressing its impact on BLT prevention and treatment remain limited due to the rarity of this tumor [10,11].

OBSERVATION

A 45-year-old man with no significant past medical history presented with a progressively enlarging perineal tumor that had been evolving over a five-year period. The lesion was associated with occasional discomfort but no systemic symptoms. Dermatological examination revealed multiple exophytic, vegetative, darkly pigmented

tumors involving the penile shaft, glans and scrotum. The lesions ranged from 1 to 10 cm in diameter and exhibited a characteristic cauliflower-like, papillomatous surface consistent with a giant condylomatous proliferation **(Figure 1)**.

Figure 1: Multiple Exophytic, Vegetative, Blackish Tumors

The dermoscopic pattern combined finger like papillomatous structures, keratin-rich surfaces and glomerular vascular patterns on a reddish-brown background **(Figure 2)**.

Figure 2: Dermoscopy Showing Filiform and Round Projections, With Dendritic Blood Vessels

Given the clinical presentation, an excisional biopsy was performed under local anaesthesia. Histopathological examination confirmed the diagnosis of Buschke–Löwenstein tumor. The specimen corresponded to cutaneous tissue containing a predominantly exophytic lesion with focal endophytic components. The lesion was lined by a markedly hyperplastic, papillomatous epidermis forming epithelial stromal vegetations with a fibrovascular core that was variably slender or abundant and well vascularized. The epidermis showed pronounced hyperkeratosis and parakeratosis, with associated koilocytic vacuolization, binucleated keratinocytes and occasional dyskeratotic cells, predominantly located within the superficial epidermal layers **(Figures 3-6)**.

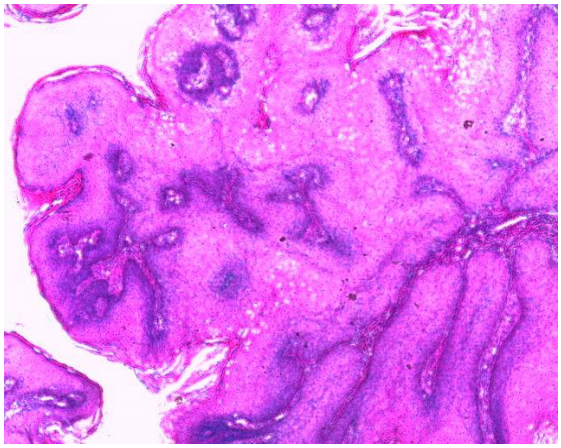

Figure 3: HES G X 50 -> Epithelio-Connective Vegetations, Hyperkeratosis + Parakeratosis + Koilocytes

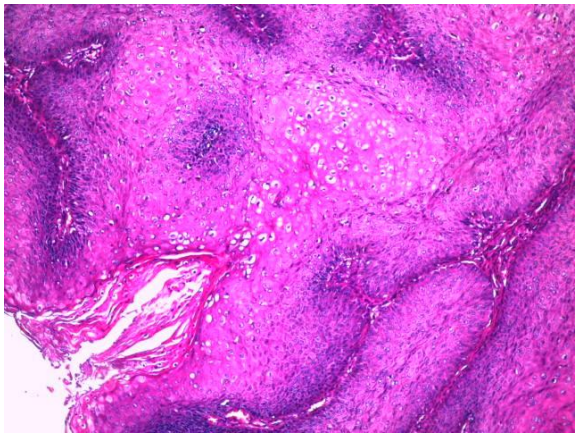

FIGURE 4. HES G X 100 -> Epithelio-Connective Vegetations, Hyperkeratosis + Parakeratosis + Koilocytes

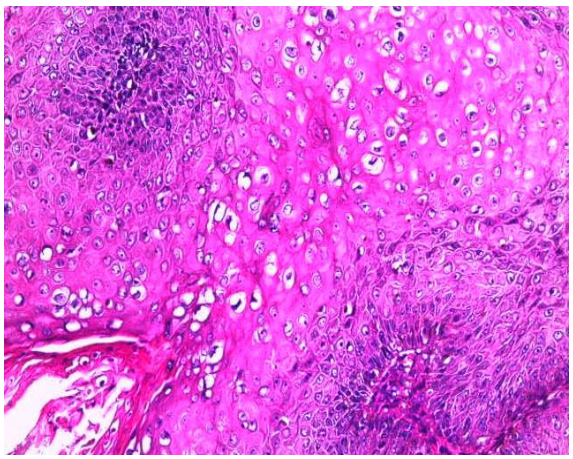

Figure 5: HES G X 200-> Details of Koilocytes + Parakeratosis

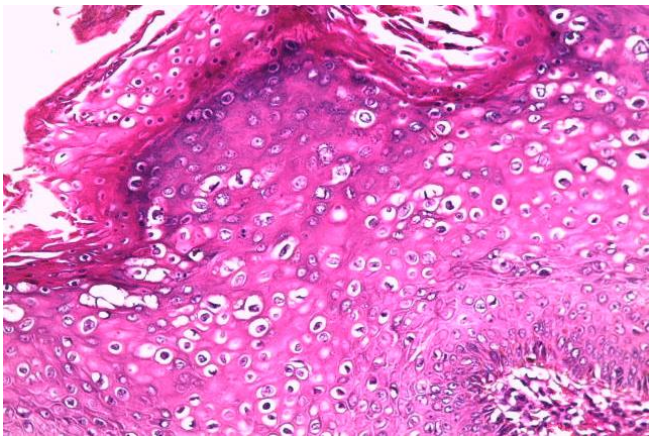

Figure 6: HES G X 200-> Details of Koilocytes + Parakeratosis

In addition, the papillomatous epidermis formed elongated, bulbous rete ridges with focal endophytic extensions, some of which were anastomosing and extending into an edematous fibrous dermis. The underlying dermis contained congested capillary vessels and a moderate mononuclear inflammatory infiltrate. Importantly, no cytonuclear atypia, abnormal mitotic figures, dysplasia or malignant transformation were identified. The basement membrane remained intact throughout the lesion and the adnexal structures and hypodermis were unremarkable.

Following histological confirmation, wide surgical excision under local anaesthesia was undertaken, resulting in a reduction of more than 80% of the tumor burden. Given the extent of the lesions and to minimize morbidity, adjuvant treatment was initiated immediately after surgery with repeated sessions of chemical destruction using 50% trichloroacetic acid. Four months later, topical immunomodulatory therapy with imiquimod was introduced as a maintenance and consolidation treatment.

After one year of clinical follow-up, the patient demonstrated satisfactory healing with excellent local control and no evidence of clinical recurrence **(Figures 7,8)**.

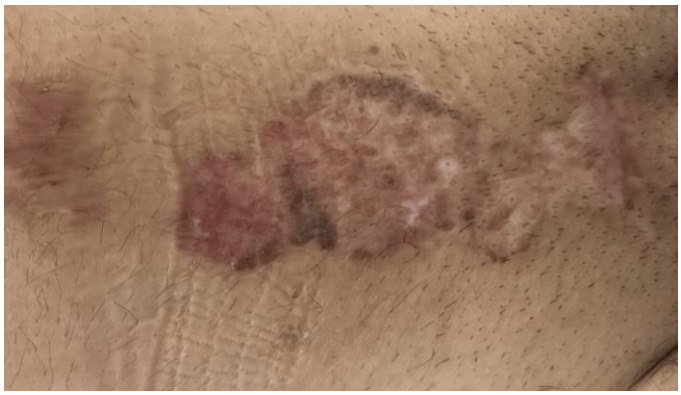

Figure 7: Six Months Follow Up

Figure 8: One Year Follow Up

This favorable outcome highlights the effectiveness of a combined therapeutic approach involving surgical debulking under local anesthesia followed by staged adjuvant topical treatments in the management of Buschke-Löwenstein tumor.

DISCUSSION

Buschke Löwenstein tumor (BLT), also referred to as giant condyloma acuminatum or verrucous carcinoma, is a rare anogenital entity characterized by slow but relentless growth, marked local invasiveness and a paradoxical discrepancy between its benign histological appearance and aggressive clinical behaviour. Although distant metastases are exceptional, BLT is associated with high recurrence rates and a substantial risk of malignant transformation, reported in up to 50–56% of cases in some series [8]. These features make BLT a challenging condition to diagnose and manage, often requiring prolonged surveillance and individualized therapeutic strategies.

BLT is strongly associated with human papillomavirus (HPV) infection, particularly low-risk genotypes 6 and 11, although other genotypes have been implicated [8]. The disease typically affects adults in the fourth to sixth decades of life and shows a clear male predominance. Nevertheless, cases have been described in pediatric patients and pregnant women, highlighting the broad clinical spectrum. Identified risk factors include multiple sexual partners, chronic genital infections, poor hygiene, smoking, diabetes and immunosuppressive conditions. These associations emphasize the importance of preventive strategies, patient education and early screening, particularly in vulnerable populations [5,6].

Clinically, BLT usually presents as a large, exophytic, cauliflower-like mass involving the genital, perineal or perianal regions. Symptoms may range from local discomfort, bleeding and infection to obstructive manifestations affecting defecation or urination. Due to its indolent growth, diagnosis is often delayed, allowing lesions to reach considerable size and increasing the risk of complications. Histopathological examination

remains essential for diagnosis, as it allows differentiation from conventional squamous cell carcinoma. Typical features include papillomatosis, acanthosis, koilocytic changes reflecting HPV infection, broad pushing rete ridges and preservation of the basement membrane, without stromal invasion or significant cytological atypia.

Surgical excision with clear margins is widely regarded as the cornerstone and gold standard of BLT management [12-14]. Several studies have demonstrated that negative surgical margins significantly reduce recurrence rates, whereas incomplete excision is associated with a high likelihood of relapse. Tumor size has also been identified as an important prognostic factor, with larger lesions correlating with increased recurrence and morbidity. The primary objective of surgery is complete tumor eradication while preserving function, particularly in anatomically sensitive regions such as the genital and anorectal area [14].

In extensive lesions, surgical management can be technically demanding and may necessitate reconstructive procedures to restore anatomical integrity and function. A wide range of reconstructive techniques has been described, including local and regional flaps, skin grafts and, in selected cases, more complex myocutaneous flaps. Abdominoperineal resection is reserved for exceptional situations involving anorectal invasion or malignant transformation. While fecal diversion via colostomy may facilitate wound healing in selected cases, it is associated with additional morbidity and should be considered cautiously, taking into account patient-related factors and disease extent [14].

Non-surgical and adjuvant therapies including topical agents (podophyllin, imiquimod), intralesional or topical 5-fluorouracil, interferon, cryotherapy, laser ablation, photodynamic therapy, chemotherapy and radiotherapy have been reported with variable success [14]. When used as monotherapy, these approaches are generally associated with high recurrence rates. However, in selected patients or as adjuncts to surgery, combined therapeutic strategies may help reduce tumor burden, improve local control and preserve function. The absence of standardized treatment guidelines underscores the need for an individualized approach based on tumor characteristics, patient comorbidities, immune status and available resources.

In the present case, surgical debulking under local anaesthesia achieved substantial tumour reduction, followed by adjuvant chemical destruction with trichloroacetic acid and delayed introduction of topical imiquimod. This combined approach allowed effective local disease control while minimizing surgical morbidity. At one year of follow-up, no recurrence was observed, supporting the role of staged, multimodal therapy in selected cases of BLT.

Given the high recurrence rates reported in the literature reaching up to two-thirds of cases in some series long-term follow-up is mandatory, even in the absence of histological malignancy. Surveillance protocols should include regular clinical examinations over several years to enable early detection of recurrence or malignant transformation. Ultimately, the management of BLT benefits from a multidisciplinary approach involving dermatology, surgery, pathology, oncology and preventive medicine. Early recognition, appropriate surgical

planning and tailored adjuvant therapy are essential to improve outcomes, reduce morbidity and preserve quality of life in patients affected by this rare but clinically significant disease [15].

CONCLUSION

Buschke–Löwenstein tumor is a rare but clinically important anogenital condition, characterized by a deceptively benign histological appearance combined with aggressive local behavior, a high risk of recurrence and potential malignant transformation. Early recognition, supported by accurate histopathological confirmation, is essential to guide appropriate therapeutic decision-making and improve outcomes. Surgical excision with clear margins remains the cornerstone of management and represents the most effective treatment strategy, particularly when combined with immediate reconstruction to preserve function and reduce morbidity. Owing to the technical complexity of resection and reconstruction, management often requires a multidisciplinary approach involving specialized surgical expertise. Although various non-surgical therapies have been described, their effectiveness remains limited. Careful preoperative assessment, individualized surgical planning and long-term follow-up are crucial to minimize recurrence and optimize oncologic, functional and cosmetic results.

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