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Case Report

A Rare Genital Tumor in a Patient with Immunosuppression: A Case Report of a Bushcke-Lowenstein Tumor

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Abstract: The Buschke-Lowestein tumor, also known as giant condyloma acuminatum, is a neoplasm affecting the anogenital region. It is caused by human papillomavirus (HPV). The lesion is characteristically slow-growing and warts-like, with a propensity for occurrence in the anogenital region. The preferred management approach is surgical resection. However, there is ongoing debate regarding the optimal use of radiotherapy, systemic chemotherapy, and topical agents.

Keywords: Condyloma Giant, Condyloma acuminatum, Buschke-Lowenstein, immunosuppression, Human papillomavirus.

INTRODUCTION

Giant condyloma acuminatum, also known as Buschke-Lowestein tumor, is an uncommon neoplasm that develops from pre-existing warty lesions. The tumor was first described by Abraham Buschke and Ludwig Löwenstein in 1925 [1,2].

It is associated with human papillomavirus (HPV) infections, with serotypes 6 and 11 being the most identified, serotypes 16 and 18 are less frequently detected. Additional risk factors include immunosuppression, human immunodeficiency virus (HIV) infections, and multiple sexual partners [3].

It is a slow-growing tumor with locally destructive and aggressive behavior, presenting as a large, cauliflower-shaped growth and exhibiting ulceration and necrotic areas [1].

The tumor does not exhibit histological evidence of malignancy, such as basement membrane infiltration, lymphatic invasion, hematogenous or distant metastasis [4].

The most described therapeutic approach is surgical excision, with a reported success rate of 90% [3]. This is often combined with adjuvant therapy using topical agents, including podophyllin, 5-fluorouracil, bleomycin, interferon, and imiquimod. In some cases, systemic therapy with chemotherapy or radiotherapy may also be employed [1]. Some authors posit that it is a low-grade squamous cell carcinoma, while others suggest that it represents a transitional form between condyloma acuminatum and squamous cell carcinoma. Malignancy rates of 30 to 56% have been reported in the literature [4, 5].

The differential diagnosis includes Bowen's disease in its condylomatous form, pseudoepitheliomatous balanitis caused by fungi, spinocellular epitheliomas, warty carcinomas, syphilis, and lymphogranulomatosis venereal [5].

CASE PRESENTATION

A 62-year-old Mexican woman with a history of diabetes mellitus, hypothyroidism, and non-Hodgkin's lymphoma that has persisted for a year and is currently undergoing treatment with chemotherapy involving cyclophosphamide, doxorubicin, and vincristine. She has not received the HPV vaccine.

The dermatosis presents as a localized condition, exhibiting a propensity for symmetry within the trunk, genitourinary region—particularly the labia minor, majora, and perianal area—and manifesting as wart-like neoformations. The lesions resemble bunches of grapes and are characterized by new neoformations of similar characteristics that are confluent and settled upon each other, with residual hemorrhagic material on their surface. These lesions measure 10*4*3 cm with well-defined borders and are situated on a base of healthy skin and mucosa.

The patient also reported experiencing pain (rated 7/10 on the visual analog scale), pruritus, and scanty bleeding in the lesion.

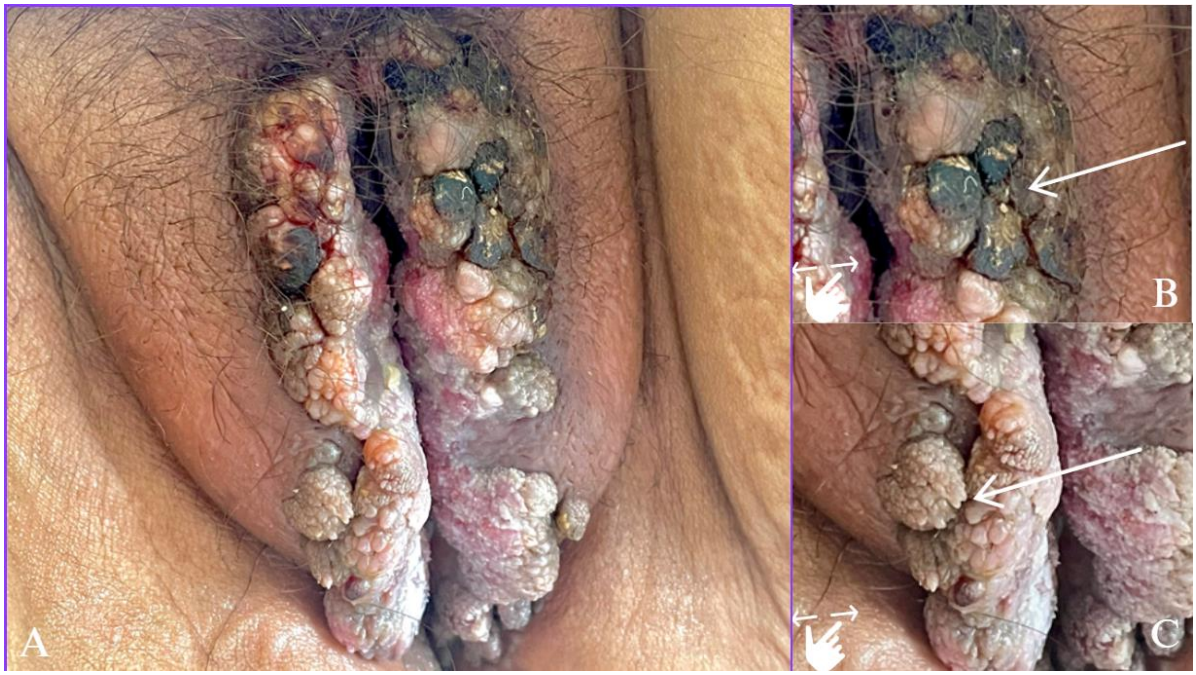

Figure A: Extensive condyloma in the vulvar region, affecting both the labia majora and minora; Figure B: A close-up of figure A reveals the presence of crusts and slight bleeding; Figure C: A close-up shows the characteristic "cauliflower" shape of these projections

Laboratory tests yielded the following results: The hemoglobin level was recorded as 8.70 mg/dl, hematocrit level 27.5%, leukocyte count 4.65 10^9/L, neutrophil count 3 10^9/L, platelet 235 10^9/L, glucose 237 mg/dl and creatinine 0.48 mg/dl. The viral panel for hepatitis B, hepatitis C, and HIV yielded negative results.

The patient was assessed by the internal medicine and gynecology departments and was scheduled for resection of the lesion with margins by the dysplasia service. However, the patient did not attend the scheduled surgery and lost follow-up.

DISCUSSION

Buschke-Lowestein tumors or giant acuminate carcinomas are rare entities that may go unnoticed due to a focus on other diseases, a lack of initial symptoms, taboo subjects related to religion or social reasons, and other factors.

The correlation between this disease and the human papillomavirus (HPV), particularly serotypes 6, 11, and less commonly 16 and 18, underscores the significance of vaccination for the prevention of not only this entity but predominantly cervical cancer [3]. The patient had not received the vaccine since the vaccination schedule was not included in the free national vaccination booklet. Furthermore, she was unaware that a vaccine was available privately.

The age range of presentation, described in the literature as occurring between the fourth and sixth decades of life, coincides with the patient's initial diagnosis of lymphoma, which was received at the onset of the lesion. Additionally, there is an established relationship between this tumor and immunosuppression; therefore, it's important to consider the possibility of other conditions that may present similarly, such as immunosuppression due to HIV, hepatitis, secondary syphilis, inguinal granuloma, tuberculosis, Nicolas Favre's disease, warty carcinoma, and squamous cell carcinoma [1,4,5].

The lesion is a slow, exophytic growth in the form of cauliflower that typically has a mass effect. It may also present as a perianal abscess or fistula due to its propensity to infiltrate adjacent tissues, affecting the perianal region and the anus. The lesions were observed in the vulva, vagina, scrotum, perineum, and bladder. The patient also reported discomfort when sitting due to the size of the lesion, pruritus, and sometimes slight bleeding when rubbing against underwear. These symptoms are also described in clinical cases [4, 5].

A histopathological report is unavailable for the patient. However, the expected findings are hyperkeratosis, acanthosis, and basal hyperplasia. Additionally, there will be irregular nuclei with vacuolated cytoplasm. The characteristics that differentiate this tumor from squamous cell carcinoma include a thickened stratum corneum and a tendency to deep invasion; however, there is an absence of basement membrane infiltration, as well as no evidence of lymphatic or vascular involvement, metastasis, dysplasia, or atypia. These factors allow for the differentiation of this type of tumor from other neoplasms [5, 6].

Imaging studies should be performed in patients with this tumor to delimit the extent of its infiltration [6, 7].

While there is no general agreement regarding the optimal therapeutic strategy, surgical excision with wide margins has demonstrated superior outcomes, with a reported success rate of 90%. It is important to acknowledge, however, that recurrence rates may reach 66%, and that the wound healing process may be affected due to the proximity to the anal sphincter. The potential benefits of chemotherapy and radiotherapy have been considered, but their use is controversial due to the lack of long-term results and the appearance of new condylomas. Additionally, our patient was already undergoing chemotherapy for Hodgkin's lymphoma, which precluded the use of these therapeutic options [2, 7].

The evidence regarding the efficacy of topical treatments is inconclusive. For instance, Podophyllin yielded no discernible benefit. Interferon has been associated with high recurrence rates, although it has demonstrated eradication rates between 45 and 60%. Imiquimod has been shown to result in complete regression of lesions. Therefore, a personalized approach is recommended for each case, with long-term follow-ups due to high recurrence rates [2, 7].

CONCLUSIONS

It is possible that genital region diseases may not be manifested by patients who present other diseases for various reasons. An adequate comprehensive assessment could lead to the diagnosis, treatment, and follow-up of rare diseases such as Buschke-Löwenstein tumor. This is mostly related to immuno-compromise, is typically benign, but there have been reports of malignant transformations and recurrence following treatment.

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