

CASE REPORTS

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Buschke–Löwenstein tumor of the penis

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Abstract

Background: The Buschke–Löwenstein tumor comes from the confluence of multiple condyloma acuminata and is clinically manifested by warty, exophytic, ulcerated lesions, with aggressive behavior, rapid growth, invasion and destruction of adjacent structures.

Case presentation: A 57-year-old man with type II diabetes mellitus, high blood pressure and a history of high-risk sexual behavior with multiple partners was evaluated in the urology department for multiple penile lesions of verrucous appearance and fetid odor of 10 months of evolution. Biopsy of the lesion was performed revealing a giant condyloma acuminatum.

Conclusions: Radical surgical excision with wide surgical margins remains the first line of treatment. Close follow-up of these patients is crucial given the complexity and tumor recurrence.

Keywords: Buschke–Löwenstein, Penis, Tumor

1 Background

The Buschke–Löwenstein tumor is an epithelial tumor initially described in 1925. It tends to present in the fifth decade of life with a male-to-female ratio of 2.7:1. Data show that approximately 3–4 million cases of genital warts in men occur each year with a peak rate of 500 per 100,000 in the 25–29-year-old age group [1]. There is an etiological relationship with the human papillomavirus (HPV). More than 30 genotypes of HPV can infect the genital epithelium. Genital warts due to HPV are mainly associated with genotypes 6 and 11 (low risk), while anogenital squamous cell carcinoma is associated with genotypes 16, 18, 31 and 33 (high risk). Multiple investigations have been conducted to differentiate verrucous carcinoma (VC) from giant condyloma of Buschke–Löwenstein (GCBL) and its relation to HPV2 [2]. Risk factors include lack of personal hygiene, promiscuity, smoking, chronic irritation, presence of foreskin and immunodeficiency (congenital and acquired) [3]. The tumor comes from the confluence of multiple condyloma acuminata

and is clinically manifested by warty, exophytic, ulcerated lesions, with aggressive behavior, rapid growth, invasion and destruction of adjacent structures. In men, it is usually located in the penis (81–94%), anorectal region (10–17%) and urethra (5%), but it can be found in the scrotum and bladder [4]. In the penis, lesions typically occur around the coronal sulcus, frenulum and glans. Despite benign histology, the malignant transformation of Buschke–Löwenstein tumor has been reported in 30–56% of cases [5–7].

2 Case presentation

A 57-year-old man with type II diabetes mellitus, high blood pressure and a history of high-risk sexual behavior with multiple partners was evaluated in the urology department for multiple penile lesions of verrucous appearance and fetid odor of 10 months of evolution, which prevented the foreskin retraction. Physical examination revealed several confluent penile lesions of exophytic morphology including the prepuce, glans and coronal sulcus (Fig. 1). Serologic tests were negative for various sexually transmitted infections, including human immunodeficiency virus (HIV) infection. Along with the physical examination, an abdomino-pelvic CT scan was used to demonstrate the location and extension of the lesion as well as the involvement of inguinal lymph

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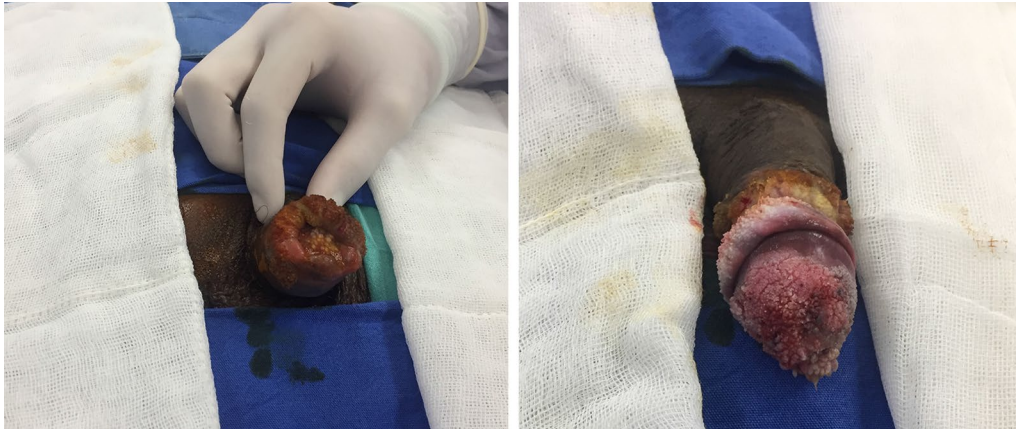


Fig. 1 Penile lesion of verrucous morphology including the prepuce, glans and coronal sulcus

nodes, which were not affected. Biopsy of the lesion was performed revealing a giant condyloma acuminatum associated with HPV. Conservative treatment with imiquimod cream (5%) was administered 5 days a week for a period of 1 month without any effective response, for which a partial penectomy was performed (Figs. 2, 3). Management of the neourethra was crucial, being dissected from the corpus spongiosum distally for a distance of approximately 1 cm and transected. It was spatulated on its dorsal surface to facilitate reconstruction and prevent stenosis. The spatulated urethra was approximated to the penile skin to create an oblique meatus with its open side at the 12 o'clock position. The length of the penile shaft remained after the surgical procedure was approximately 3 cm, considered an acceptable length to maximize functional outcomes. At follow-up, the patient preserves the ability to void in the standing position with effective capacity to direct the urinary stream. Sexual function was maintained due to a penis length greater than 2 cm, considered the minimum length to allow intercourse. The patient remains with satisfactory evolution in close surveillance due to the high risk

of recurrence, with consultations at our outpatient clinic every 3 months during the first year in which physical examination of the penis, urethral meatus and inguinal region is performed in search of new lesions, stenosis and adenopathies.

3 Discussion

GCBL has been treated by several modalities, which can be classified into three types: topical therapy (e.g., podophyllin, fluorouracil or radiotherapy), tumor removal (e.g., cryotherapy, CO₂ laser therapy or surgical excision) and immunotherapy (e.g., imiquimod). However, the variety of treatment regimens currently applied does not allow the formulation of definitive therapeutic guidelines. The application of imiquimod (5%), podophyllin (25–30%), trichloroacetic acid, 5-fluorouracil and bleomycin alone or combined with cisplatin or methotrexate has had variable results. Treatment with interferons 2 α and 2 β combined with laser therapy (Nd:YAG) has been described, as well as cryosurgery with some cases of success. Radical surgical excision with wide surgical margins remains the first line of treatment, with a higher success

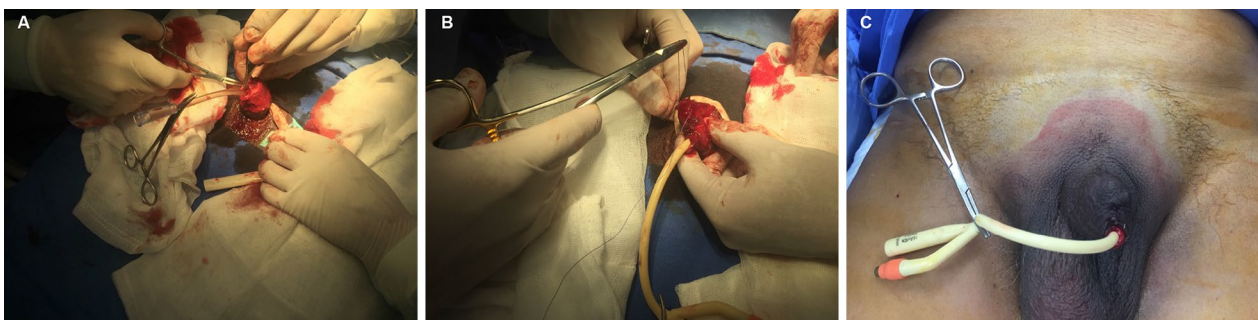


Fig. 2 Partial penectomy. **a** Urethral repair. **b** Closure of cavernous bodies. **c** Remaining penile tissue

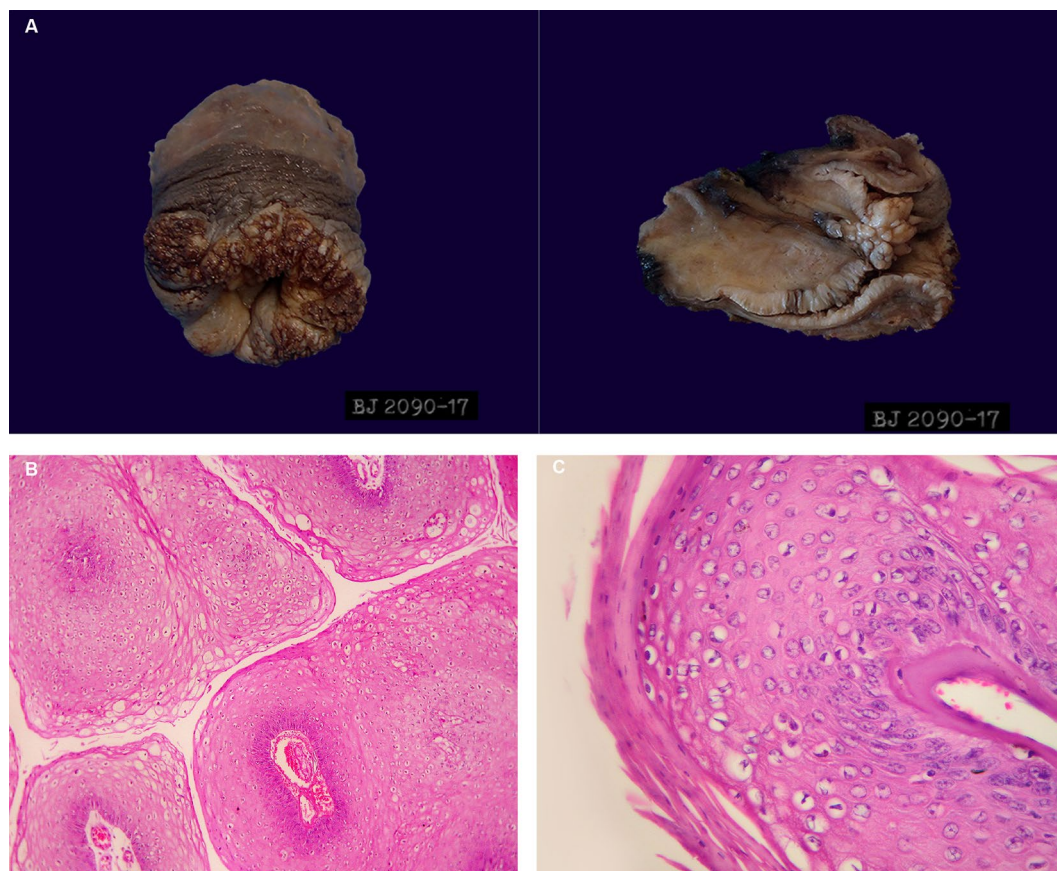


Fig. 3 a Macroscopic product of partial phallectomy. b Papillary projections coated by stratified squamous epithelium with acanthosis. c Cytopathic changes associated with human papillomavirus (HPV)

rate (63–91%) and a lower rate of relapse [8]. Nevertheless, penile skin loss and repair of the urethra can be a challenge for urologists. Surgical options include one-stage or two-stage procedures, using either single or multiple tissue transfer [9, 10]. Preputial and various penile skin flaps, such as a longitudinal flap, the hockey stick flap, the penile island flap and the circumferential/circumpenile flap, have been used for penile urethral reconstruction as good options when needed [11, 12]. A large representative biopsy specimen is important to judge the structure of the lesion in order to establish the diagnosis and to exclude VC [13]. In microscopic examination, invasion of the subepithelial tissue is seen by expansion rather than by infiltration, leaving the basement membrane intact. A well-stratified epithelium is shown with minimal cellular dysplasia or atypical cells, rare mitotic figures, acanthosis and hyperkeratosis and no evidence of neural or vascular invasion [14]. The risk of recurrence after excision is 60–66%, with an overall mortality of

20–30% [15]. Close follow-up of these patients is crucial given the complexity and tumor recurrence.

4 Conclusions

The Buschke–Löwenstein tumor is a rare disease characterized by giant slow growing condyloma acuminatum that is locally aggressive and destructive. Although there have been reports of successful treatment with conservative modalities, the only consistently effective therapy is wide surgical excision of the tumor with clear margins with or without adjuvant chemotherapy.

Abbreviations

HPV: human papillomavirus; VC: verrucous carcinoma; GCBL: giant condyloma of Buschke–Löwenstein; HIV: human immunodeficiency virus; CT: computerized tomography; CO₂: carbon dioxide; Nd:YAG: neodymium-doped yttrium aluminum garnet.

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Authors' contributions

JP researched the literature and wrote the manuscript. JP and GM operated on the patient and had the idea for this case report. JL checked the manuscript and made corrections. MS contributed with the histopathology report and histopathology images. JT and CV provided the overall guidance and support. All authors read and approved the final manuscript.

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Availability of data and materials

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

Ethics approval and consent to participate

The study was approved by the Research Ethics Committee of the Department of Education of the Juárez Hospital of Mexico.

Consent for publication

Written informed consent was obtained from the patient for their anonymized information to be published in this article.

Competing interests

The authors declare that they have no competing interests.

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