OXFORD

Oxford Medical Case Reports, 2024, **4**, 160–162
https://doi.org/10.1093/omcr/omae034
Case Report

Buschke-Lowenstein tumor in the penis and anorectal region: case report

Badr Kabila[1,*], Kaoutar Imrani[1], Soufiane Kaddouri[1], Ismail Boujida[2], Fouad Zouaidia[2], Nabil Moatassim Billah[1] and Itimad Nassar[1]

[1]Radiology Central Department, Ibn Sina University Hospital, Mohammed V University, Rabat, Morocco
[2]Anapatology Department, Ibn Sina University Hospital, Mohammed V University, Rabat, Morocco

[*]Correspondence address. Avenue Hassan 2, Rabat, Morocco. Tel: +212 614453732; E-mail: drkabila49@gmail.com

Abstract

The Buschke-Lowenstein Tumor is a giant condyloma acuminatum caused by human papillomavirus, most commonly types 6 or 11. It is a rare condition with an estimated frequency of 0.1% in the general population. Transmission primarily occurs through sexual contact. It particularly affects men, predominantly appearing on the penis, characterized by its deep-seated growth, potential for degeneration, and tendency to recur after treatment. Surgery is the preferred treatment. We present the case of a50-year-old patient with a history of recurrent urethritis and multiple sexual partners. The patient sought medical attention for a swelling on the penis that had been progressively developing over the past 10 years. An MRI was performed for a locoregional study, revealing a perineal and penile mass with polylobed contours resembling a cauliflower, along with a sizable perineal mass Histological examination of a biopsy sample from the penis confirmed the diagnosis. Unfortunately, due to the tumor's characteristics and the patient's condition, surgery was contraindicated.

INTRODUCTION

The Buschke-Lowenstein Tumor (BLT) is a rare clinical entity of viral origin, primarily transmitted through sexual contact. Its severity is attributed to the risk of local invasion, strong tendency to recur, and the potential for degeneration.

CASE REPPORT

A 50-year-old patient with a history of recurrent urethritis and multiple sexual partners presented with a swelling of the penis that had progressively developed over 10 years. The examination revealed a massive swelling of the penis, appearing grayish and pinkish, exhibiting a papillomatous, irregular, cauliflower-like appearance (Fig. 1). Multiple inguinal lymphadenopathies were observed, accompanied by a general deterioration of health. HIV, syphilis, and hepatitis serologies were negative. An MRI was requested for a locoregional study, revealing a perineal and penile protruding process with lobulated contours surrounding the penis, resembling a cauliflower appearance in intermediate T2 signal, with diffusion restriction and enhancement after Gadolinium injection, remaining at a distance from the corpora cavernosa and the scrotum (Fig. 2). Additionally, a sizable perineal mass in hypo-intense T2 signal, discreetly enhanced after Gadolinium injection, infiltrating the internal and external sphincters and extending to the scrotum, was noted (Fig. 3).

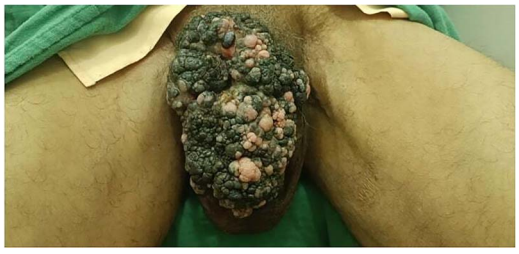

Figure 1. Grayish and pinkish lesions, presenting a papillomatous, irregular, cauliflower-like appearance on the penis.

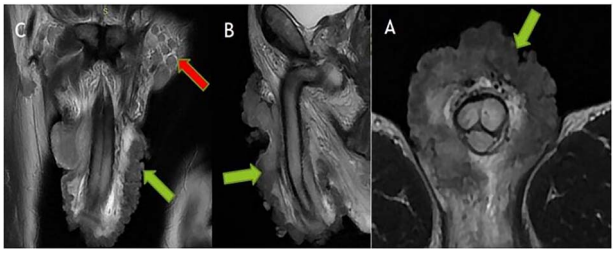

Figure 2. MRI of the penis (A: Axial T2 sequence, B: Sagittal T2, C: Coronal T2) showing a perineal and penile protruding process with lobulated contours surrounding the penis, resembling a cauliflower appearance in intermediate T2 signal (Arrow).

Received: January 16, 2024. **Revised:** February 26, 2024. **Accepted:** March 8, 2024

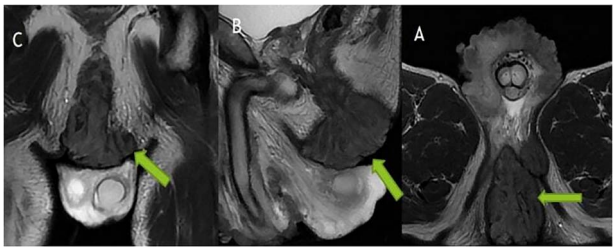

Figure 3. MRI of the penis (A: Axial T2 sequence, B: Sagittal T2, C: Coronal T2) revealing a perineal mass with hypo-intense T2 signal, infiltrating the internal and external sphincters, extending down to the scrotum.

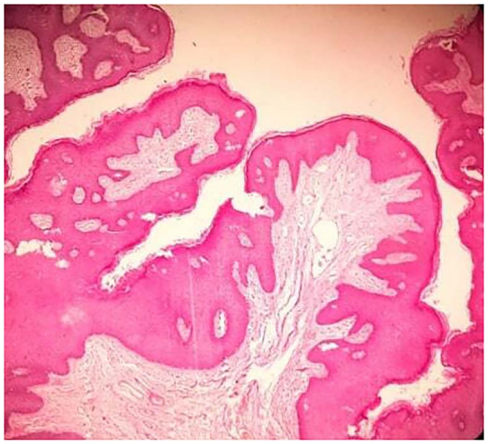

Figure 4. Histology—Lining of acanthotic and papillomatous squamous cells, topped with parakeratotic hyperkeratosis, and the presence of koilocytes.

The histological examination of a biopsy sample from the penis revealed an epitheliomatous hyperplasia composed of a layer of acanthotic and papillomatous squamous cells, topped with parakeratotic hyperkeratosis. The presence of koilocytes indicated an HPV infection, confirming the diagnosis of Buschke-Löwenstein Tumor, without signs of malignancy, the inguinal adenopathies are therefore of infectious origin and not metastatic (Fig. 4). Unfortunately, considering the extent of the tumor and the patient's overall condition, surgery was deemed contraindicated, and the chosen approach will be radiochemotherapy. After one month, clinical stability was observed.

DISCUSSION

The Buschke-Löwenstein tumor is a rare sexually transmitted disease that affects the anogenital region, caused by the human papillomavirus, particularly genotypes 6 or 11 [1]. It typically occurs in men with a history of recurrent and treatment-resistant anogenital warts. Its frequency is estimated to be 0.1% of the general population [1] and was first described in 1896. Buschke and Löwenstein officially characterized it as a distinct entity in 1925 [2].

This tumor can occur at any age after puberty, with an average onset around 45 years, and it exhibits a higher prevalence in males [2].

In men, Buschke-Löwenstein tumor is primarily localized to the external genital organs, with a predominant occurrence on the penis. In rare cases, it may also affect the ano-rectal region [3]. For women, the tumor is mainly vulvar in 90% of cases, with ano-rectal localization being less common [3].

Immunosuppression, multiple sexual partners, engagement in prostitution, homosexuality, poor hygiene, and chronic infections contribute to HPV transmission. Chronic inflammation appears to be a risk factor for this condition [4].

The transmission of Buschke-Löwenstein Tumor primarily occurs through sexual contact and also through contaminated water, clothing, gloves, and other soiled materials. The virus is resistant to environmental conditions [4].

The tumor is always preceded by grayish or pinkish condylomatous lesions, progressively evolving to take on a papillomatous, irregular, cauliflower-like appearance [5]. It develops both on the surface and in depth, distinguishing it from ordinary acuminated condylomas. The extension can reach the scrotum or vulva, the intergluteal cleft, buttocks, and even the rectum and pelvis. On the surface, it can give rise to a massive tumor measuring several centimeters. In depth, the tumor evolves by destroying and displacing nearby structures without infiltrating them [5]. The presence of bleeding, infiltration of the base, or the presence of lymphadenopathy should raise suspicion of malignant degeneration.

The patient sometimes seeks medical attention for peri-anal pain, itching, rectal bleeding, purulent discharge, weight loss, and/or the palpation of a perineal mass. A comprehensive clinical examination, including palpation of the lymph nodes, is necessary. Locoregional lymphadenopathy is often found due to frequent superinfection of the lesions.

The Buschke-Lowenstein tumor rarely metastasizes to the inguinal lymph nodes, inguinal lymphadenopathy may develop as a result of inflammation or infection associated with the tumor. Additionally, if the tumor progresses and spreads locally, it may involve nearby lymph nodes, leading to lymphadenopathy.

Overall, while inguinal lymphadenopathy can be seen with a variety of genital conditions, its presence doesn't necessarily imply a specific relationship with Buschke-Lowenstein tumor unless there is direct involvement or spread of the tumor to the lymph nodes [5].

Locoregional imaging (Ultrasound, CT scan and/or magnetic resonance imaging) is required to assess perineal tumor extension and the operability of the lesion.

Computed tomography and especially Magnetic Resonance Imaging play a crucial role in patients at high risk of invasive disease and malignant transformation. When assessing the CT imaging of such lesions, it is essential to focus on determining the degree of local infiltration and invasion into subcutaneous tissue, muscles, pelvic cavity, genital organs, rectum, and perineum. Additionally, CT scans can provide valuable insights into the considerable vascularization of these lesions. (MRI) could also be employed to assess condyloma acuminatum, providing additional details on soft tissues and aiding in surgical planning. Furthermore, imaging may also play a role in follow-up evaluations to assess recurrence [6].

Finally, a comprehensive assessment of other sexually transmitted infections will be conducted, including HIV serology, venereal disease research laboratory-*Treponema pallidum* hemagglutination assay (VDRL-TPHA), Chlamydia polymerase chain reaction (PCR), and hepatitis B serology [7].

The Buschke-Lowenstein tumor poses a challenge in differential diagnosis with other pathologies. Indeed, certain tumor (squamous cell carcinomas) or infectious lesions (secondary syphilis, verrucous and vegetative tuberculosis, Nicolau Favre disease, granuloma inguinale or Donovanosis, ano-genital amebiasis) can mimic TBL at the clinical stage [8].

Biopsy of the lesion is essential for histological confirmation and aids in identifying areas of degeneration. Under optical

microscopy, TBL presents as a clearly delineated squamous tumor, marked by considerable epithelial hyperplasia, occasionally pseudo-epitheliomatous, with an intact basal membrane, hyperacanthosis, hyperpapillomatosis, and the presence of koilocytes. Koilocytes serve as pathognomonic indicators of HPV infection, though their presence may vary [9].

Surgery remains the treatment of choice for Buschke-Löwenstein Tumor [9]. It should be sufficiently extensive to minimize the risk of recurrence and aim for a definitive cure. The surgical approach varies depending on the location. In peri-anal locations, excision with sphincter preservation and reconstruction is performed whenever possible. However, more extensive procedures, such as rectal amputation or abdominoperineal amputations, may be necessary at times [9].

Chemotherapy is primarily indicated as a preoperative measure to reduce tumor volume and decrease the aggressiveness of the surgical procedure [10].

Recurrences are a common feature of this tumor and are a direct consequence of a surgically inadequate procedure. Therefore, a comprehensive surgical approach is crucial to minimize the risk of recurrence [10].

CONCLUSION

BuschkeLöwensteinTumor is a viral-origin epithelial condylomatous proliferation with uncertain evolutionary behavior and a risk of transformation into squamous cell carcinoma. Its prevention is imperative and relies on the treatment of acuminated condylomas and the prevention of sexually transmitted diseases. Early treatment is essential, predominantly involving surgical intervention requiring extensive excision.

ACKNOWLEDGEMENTS

None.

CONFLICT OF INTEREST STATEMENT

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

FUNDING

The author(s) received no financial support for the research, authorship, and/or publication of this article.

ETHICAL APPROVAL

No ethical approval is required for de-identified single case reports based on our institutional policies.

CONSENT

Written informed consent was obtained from a legally authorized representative(s) for anonymized patient information to be published in this article.

GUARANTOR

Badr Kabila.

REFERENCES

1. Chu GY, Chang TCC, Chang CH. Buschke-Löwenstein tumor (giant condyloma acuminatum) successfully treated by topical PDT: a case report. *Dermatol Sin* 2013;**31**:94–7.
2. Njoumi N, Tarchouli M, Ratbi MB, Elochi MR, Yamoul R, Hachi H. *et al.* La tumeur de Buschke-Lowenstein anorectale: à propos de 16 cas et revue de la littérature. *Pan Afr Med J* 2013; **16**:131.
3. Gillard P, Vanhooteghem O, Richert B, De La Brassinne M. Tumeur de Buschke-Löwenstein. *Ann Dermatol Venereol* 2005;**132**: 98–9.
4. Agarwal S, Nirwal GK, Singh H. Buschke-Lowenstein tumeur du gland du pénis. *Int J Surg Case Rep* 2014;**5**:215–8.
5. Lévy A, Lebbe C. Prise en charge des tumeurs de Buschke-Löwenstein. *Ann Urol (Paris)* 2006;**40**:175–8.
6. Chae JY, Bae JH, Yoon CY, Park HS, Moon du G, Lee JG. *et al.* Female urethral condyloma causing bladder outlet obstruction. *Int Neurourol J* 2014;**18**:42–4.
7. Chao MWT, Gibbs P. Squamous cell carcinoma arising in a Giant Condyloma Acuminatum (Buschke-Lowenstein tumour). *Asian J Surg* 2005;**28**:238–40.
8. Elmejjad A, Dakir M, Tahiri M. *et al.* Le condylome acuminé géant - tumeur de Buschke Lowenstein (à propos de 3 cas). *Prog Urol* 2003;**13**:513–7.
9. Reichenbach I, Koebell A, Foliguet B, Hatier M, Mascotti J, Landes P. Tumeur de Buschke et lowenstein à propos d'un cas feminin. *J Gynecol Obstet Biol Reprod* 1995;**24**:491–5.
10. Safi F, Bekdache O, Al-Salam S, Alashari M, Mazen T, El-Salhat H. Management of peri-anal giant condyloma acuminatum—a case report and literature review. *Asian J Surg* 2013;**36**: 43–52.