

BUSCHKE-LÖWENSTEIN TUMOR: REPORT OF THREE CASES

TUMOR DE BUSCHKE-LÖWENSTEIN: RELATO DE TRÊS CASOS

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ABSTRACT

The Buschke-Löwenstein Tumor is a rare, sexually transmitted disease, triggered by human papillomavirus, specially the subtypes 6 and 11. It is characterized as a cauliflower-shape exophytic mass, slowly progressive, with high local recurrence rates and high infiltration. The main risk factor is immunosuppression. Surgical treatment is usually preferred, with or without adjuvant therapy. It has a great impact on the patients' life, impairing their life quality. We report three cases of Giant Condyloma with diverse histopathological findings with varying degrees of infiltration and papillomatosis.

Keywords: Buschke-Löwenstein tumor; Condylomata Acuminata; Papillomaviridae.

RESUMO

O tumor de Buschke-Löwenstein é uma doença rara, de transmissão sexual, associada ao papilomavírus humano, principalmente dos subtipos 6 e 11. Caracteriza-se como uma lesão exofítica, em forma de couve-flor, de progressão lenta, com alto poder de infiltração local. O seu principal fator de risco é a imunossupressão e o tratamento geralmente é cirúrgico, com ou sem terapias adjuvantes. O impacto na vida da paciente é grande, com altas taxas de recorrência após excisão cirúrgica. Relatamos 3 casos de condiloma gigante com achados histopatológicos diversos, com graus de infiltração e papilomatose variados.

Palavras-chave: Tumor de Buschke-Löwenstein; Condiloma acuminado; Papilomaviridae.

INTRODUCTION

The Buschke-Löwenstein Tumor (BLT) or Giant Condyloma was first described in 1925. It is a rare sexually transmitted disease associated with human papillomavirus (HPV), most commonly subtypes 6 and 11, observed in 90.0% of cases^(1,2). Incidence is about 0.1% in the general population, more common in underdeveloped countries, and men are the most affected (3:1). The average age of the emergence of the lesion is around 50 years⁽³⁾.

The disease's pathogenesis and natural history are not well known, but it is characterized by slow development and progression of benign exophytic tumors, cauliflower-shaped, infiltrating locally and destroying the adjacent tissue. The tumor carries high recurrence rates after surgical excision (about 60.0%)^(2,4). The main risk factors are: poor hygiene, promiscuity, alcoholism, diabetes, chronic irritation, immunosuppression, and recurrent genital Condyloma^(1,5).

The ideal treatment for BLT is still under discussion, since there is no consensus in the literature, mainly due to the rarity and lack of multicentered studies or randomized trials involving a significant number of participants. We report here cases of three patients with genital BLT and discuss its variable histopathological emergence and the difficulties of handling this rare and mutilating pathology.

CASES REPORT

Patient "A", 17 years old, single, nulliparous, menarche at the age of 15, without active sex life for three months, underwent liver transplantation for autoimmune hepatitis nearly three years. Tacrolimus,

prednisone and warfarin were being administered. Patient showed condylomatous lesions in the vulva for approximately one year, when treatment was initiated with the application of trichloroacetic acid (ATA) 85.0%. Initially, the patient showed good response to the treatment, not returning to the service for five months. A lesion progression was observed during patient's return, associated with local bleeding and itching, and difficulty to be seated. Physical examination showed a Giant Condyloma in labia majora and pubic mound, sessile, painful at manipulation, with foul odor from the content, extending to the perianal region (Figure 1). A simple vulvectomy was carried out with good healing of surgical wound. Anatomopathology of the surgical piece was compatible with Condyloma (Figure 2). Patient developed new condylomatous lesions in the first six months, and administration of ATA 85.0% was held, causing the lesions to regress.

Patient "B", 27 years old, nulliparous, single, menarche at the age of 19, without active sex life for one year, in use of immunosuppressants due to kidney transplant about five years ago. Patient presents verrucous vulvar injury for about one year, with local pain at handling. On physical examination, multiple verrucous lesions in labia expanding from the supra-clitoral region to the perianal region were observed. The excision of the lesion with electric scalpel was carried out. Histopathological analysis of the surgical part confirmed the diagnosis of BLT (Figure 3). Patient evolved with good healing, without the emergence of new lesions in a period of six months.

Patient "C", 58 years old, multiparous, widow, no sex life for about four years, poorly compensated type 1 diabetic, and smoker. The verrucous lesion in vulva, of gradual increase, arose about six years ago, associated with pruritus and local pain. Patient reported weight loss of 5 kg in one month and dysuria. The physical examination showed extensive condylomatous sessile damage, with a base expanding from the lower third of labia majora to perineal and perianal regions (Figure 4). A vulvectomy by skinning was carried out. Patient evolved with severe pain in the surgical wound, edema and drainage of serous and fetid secretion, removal of stitches with

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posterior dehiscence. Antibiotic therapy was initiated, followed by good wound healing after the second procedure. The histopathological result revealed vulvar Condyloma.

DISCUSSION

BLT is a rare clinical formation with a histological pattern similar to Condyloma Acuminata^(2,6). However, it shows a subtle difference in relation to the growth pattern, as it incorporates the adjacent stroma. This growth pattern results in local infiltration and destruction of surrounding tissue, requiring a more extensive resection.



Figure 1 – Giant Condyloma in pubic mound, labia majora and perianal region (Patient “A”).

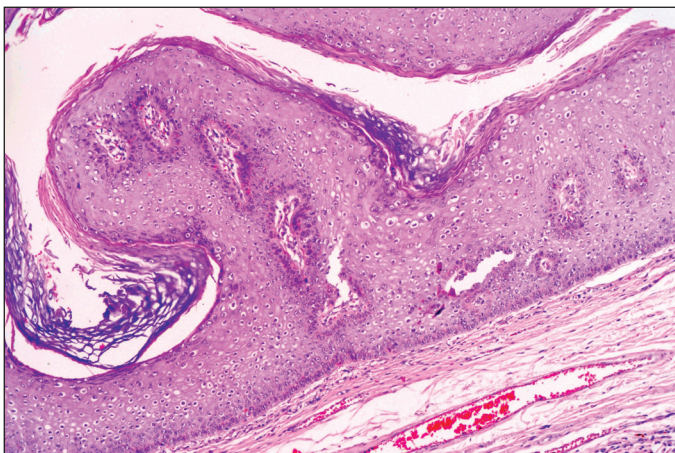


Figure 2 – Histopathology of Patient “A”: Note endophytic growth of squamous epithelium and acanthosis (H & E; X40).

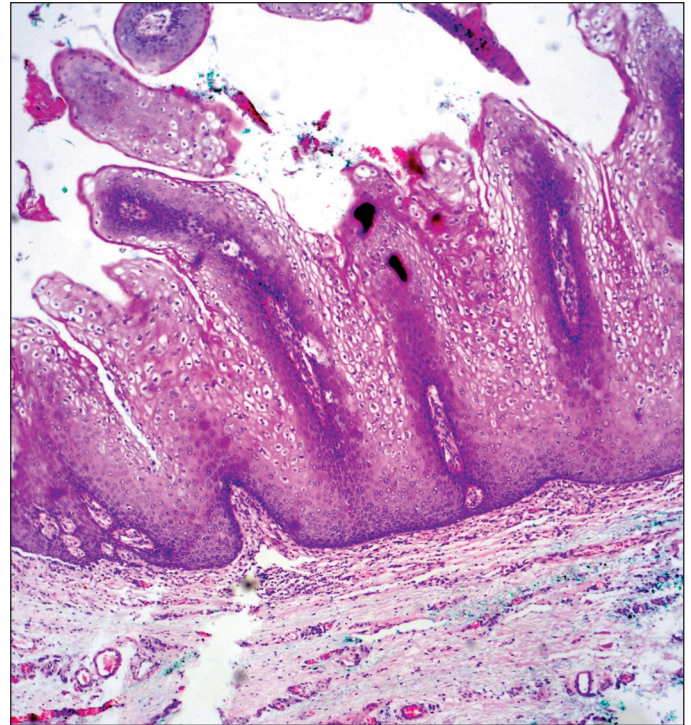


Figure 3 – Histopathology of Patient “A”: Note condylomatous epithelial surface with koilocytes (H & E; X40).

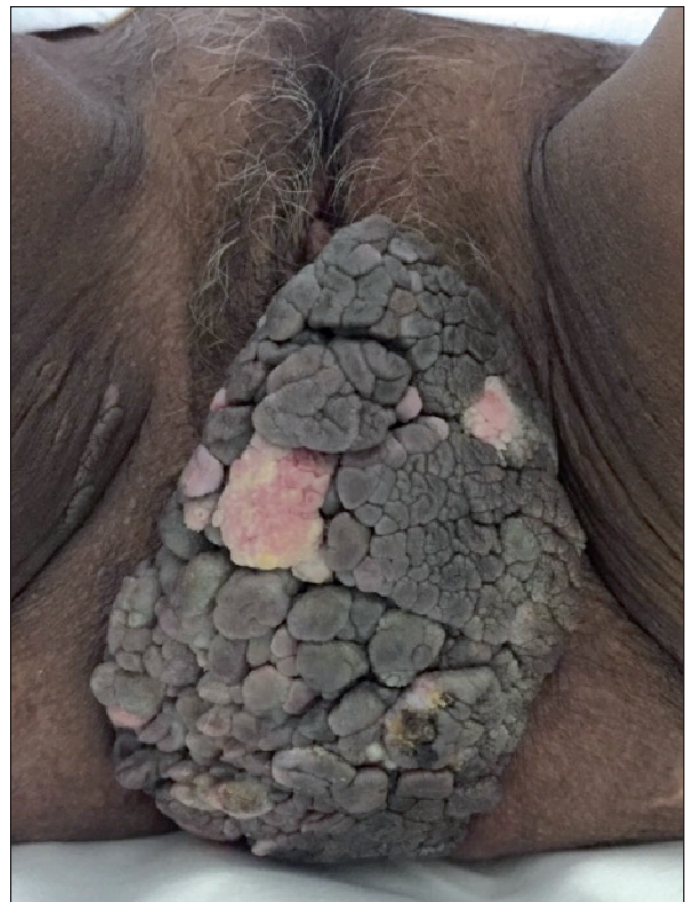


Figure 4 – Giant Condyloma of the left lower third of the labia majora until perineal region (Patient “C”).

One of BLT's features is the benign pattern that might progress to extensive papillomatous proliferation and infiltrate surrounding tissues if not properly treated⁽⁷⁾. It is estimated that 30.0–50.0% of the cases of Condyloma Acuminata have malignant transformation in Verrucous Carcinoma (VC) or squamous cell carcinoma after 5 years on average, but the increase of risk factors is not clear yet. There are no reports of remote metastasis^(8,9).

Immunocompromised patients usually present multiple lesions and multiple HPV infections, and a careful examination is required to exclude high-grade coexistent lesion^(2,6). Two of the patients underwent transplants and presented the symptom for about one year. The third one (patient "C") was diabetic and a smoker, and referred to the progressive symptom for six years. All patients reported cessation of sexual activity since the emergence of the disease, local pain, and their extension to the perianal region. Patients showed negative serologies for sexually transmitted infections.

The expansive/infiltrating behavior observed on the histopathological examination is similar to that of well-differentiated VC, causing some controversy. VC is characterized by acanthosis and papillomatosis, such as in BLT, but does not show atypical or HPV-related lesions. It is typically associated with inflammatory diseases of the vulva or lichen simplex chronicus verrucosus. BLT, unlike VC, destroys underlying tissues due to papillary proliferative process compression, but without the growth pattern of classic infiltrating carcinoma^(10,11). One of the reported cases (patient "A") presented a more endophytic growth pattern, with less koilocytes when compared to the other two (patients "B" and "C"). This differentiation between BLT and VC can be difficult considering the histopathological study, as BLT is considered by some authors as an intermediate entity between Condyloma Acuminata and VC^(1,9).

The need for a biopsy prior to the treatment is questioned, as most authors concluded that excision is the indicated treatment, not depending on the histological type. A regular follow-up is also important to evaluate the level of the lesion infiltration, both locally and systemically, in order to determine the best type of treatment. This evaluation should be performed during the preoperative period through Computed Tomography (CT) scans and/or Magnetic Resonance Imaging (MRI) of abdomen and pelvis^(9,12).

The treatment of choice is usually surgical with a radical wound excision and free margins (20 mm), with or without adjuvant therapies. This treatment shows higher rates of success (53.0–91.0%) and lower risk of recurrence. The surgery may or may not be associated with chemotherapy^(2,4,6). Recent studies do not recommend radiotherapy for BLT, as it can generate the epithelium dedifferentiation⁽⁶⁾. Recurrent tumors usually present significant cytological anomalies and are classified as typical squamous carcinomas. Topical application of podophyllin is not indicated as there are no studies on the subject. The role of topical application of 5-fluoro-uracil is not well defined either, since it is related to poor results in cases of Giant Condyloma⁽⁷⁾. Imiquimod seems to be as effective as the surgical excision in initial anogenital lesions⁽¹³⁾. The three patients described in this study underwent surgical excision and there was no BLT recurrence following.

The overall impact in the life of the patient is huge, and could be avoided with vaccination against HPV. Many of these women end up hiding out due to shame and fear of a malignant disease. In addition,

they must adapt to the lesion between their legs, relearning how to walk, sit and sleep⁽¹⁴⁾. Therefore, it is important to provide psychological support to these patients, in addition to a long-term follow-up maintenance in order to prevent, adequately treat recurrences, and avoid malignant transformation⁽⁷⁾.

Conflict of interests

The authors declare no conflict of interests.

REFERENCES

- Ahsaini M, Tahiri Y, Tazi MF, Elammari J, Mellas S, Khallouk A, et al. Verrucous carcinoma arising in an extended giant condyloma acuminatum (Buschke-Lowenstein Tumor): a case report and review of literature. *J Med Case Rep.* 2013;7:273-7.
- Niazy F, Rostami K, Motabar AR. Giant Condyloma Acuminatum of Vulva Frustrating Treatment Challenge. *World J Plast Surg.* 2015;4(2):159-62.
- Braga JC, Nadal SR, Stiepcich M, Framil VM, Muller H. Buschke-Loewenstein Tumor: identification of HPV type 6 and 11. *Ann Bras Dermatol.* 2012;87(1):131-4.
- Spinu D, Radulescu A, Bratu O, Checherita IA, Ranetti AE, Mischianu D. Giant Condyloma Acuminatum – Buschke-Lowenstein Disease – a Literature Review. *Chirurgia (Bucur).* 2014;109(4):445-50.
- 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(1):43-52.
- Tripoli M, Cordova A, Maggi F, Moschella F. Giant condylomata (Buschke-Löwenstein Tumours): our case load in surgical treatment and review of the current therapies. *Eur Rev Med Pharmacol Sci.* 2012;16(6):747-51.
- De Toma G, Cavallaro G, Bitonti A, Polistena A, Onesti Mg, Scuderi N. Surgical Management of Perianal Giant Condyloma Acuminatum (Buschke-Löwenstein Tumor). Report of Three Cases. *Eur Surg Res.* 2006;38(4):418-22.
- Chu QD, Vezeridis MP, Libbey NP, Wanebo HJ. Giant condyloma acuminatum (Buschke-Lowenstein Tumor) of the anorectal and perianal regions. Analysis of 42 cases. *Dis Colon Rectum.* 1994;37(9):950-7.
- Virgilio E, Balducci G, Mercantini P, Tozzi F, Ziparo V, Ferri M. Perianal giant condyloma acuminatum of Buschke-Loewenstein: a carcinoma-like condyloma or a condyloma-like carcinoma? *ANZ J Surg.* 2015;85(5):394-5.
- Yorganci A, Serinsoz E, Ensari A, Sertcelik A, Ortac F. A case report of multicentric verrucous carcinoma of the female genital tract. *Gynecologic Oncology.* 2003;90(2):478-81.
- Steffen C. The men behind the eponym – Abraham Buschke and Ludwig Lowenstein: giant condyloma (Buschke-Loewenstein). *Am J Dermatopathol.* 2006;28(6):526-36.
- Correia E, Santos A. Buschke-Löwenstein Tumour: Successful Treatment with Minimally Invasive Techniques. *Case Rep Dermatol Med.* 2015;2015:1-4.
- Reusser NM, Downing C, Guidry J, Tying SK. HPV Carcinomas in Immunocompromised Patients. *J Clin Med.* 2015;4(2):260-81.
- Atkinson AL, Pursell N, Sisay A. The Giant Condyloma (Buschke-Lowenstein Tumor) in the immunocompromised patient. *Case Rep Obstet Gynecol.* 2014;2014:1-4.

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