

## Case report

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# Perianal Buschke-Lowenstein tumor: report of two cases treated with 25% podophyllin ointment

*Tumor de Buschke-Lowenstein perianal: relato de dois casos tratados com podofilina em vaselina sólida 25%*

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## ABSTRACT

Buschke-Lowenstein tumor (BLT) is an extremely rare variant of condylomata acuminata that presents a benign biological behavior and histological characteristics despite being clinically presented by large lesions. Many therapeutic approaches are available, many of which rely on extensive and mutilating surgical procedures. We present two cases of patients with BLT treated with topical podophyllin whose therapeutic responses were extremely favorable in both cases.

**Keywords:** Podophyllin; Condylomata Acuminata; Carcinoma, Verrucous; Neoadjuvant Therapy

## RESUMO

O tumor de Buschke-Lowenstein (TBL) é variante extremamente rara do condiloma acuminado que, apesar de manifestar-se clinicamente por lesões de grandes proporções, apresenta comportamento biológico e características histológicas benignas. Existem diversas abordagens terapêuticas disponíveis, muitas delas apoiando-se em abordagens cirúrgicas extensas e mutilantes. Apresentamos dois casos de pacientes com TBL tratados com podofilina tópica, cujas respostas terapêuticas foram extremamente favoráveis em ambos os casos.

**Palavras-chave:** Podofilina; Condiloma Acuminado; Carcinoma Verrucoso; Terapêutica; Terapia Neoadjuvante

## INTRODUCTION

The Buschke-Lowenstein tumor (BLT), also known as giant condyloma acuminata, is a rare variant of condyloma acuminata, comprising about 0.1% of cases.<sup>1</sup> It is caused by human papillomavirus (HPV) types 6 and 11.<sup>2</sup> The risk factors described are immunosuppression, pregnancy, alcohol and tobacco consumption, poor local hygiene, and Herpes simplex virus infection.<sup>3</sup>

Clinically, the lesion reaches massive proportions, with aggressive local characteristics, invading and causing deformity in the adjacent tissues, but without lymphatic, vascular, or neuronal invasion.<sup>4</sup> The risk of degeneration for squamous cell carcinoma (SCC) ranges from 30% to 56%. At the same time, condylomata acuminata has a risk of only 2%.<sup>5</sup> Histologically, BLT is distinguished from condylomata acuminata by its pro-

liferation and deep penetration into adjacent tissues, and from SCC, by the integrity of the basement membrane and inability to produce metastases.<sup>3</sup>

BLT has several therapeutic approaches, such as topical medications, cryosurgery, surgical excision, immunotherapy, chemotherapy, radiotherapy, and electrocoagulation. There is no consensus to guide the therapeutic decision, but the initial choice in many services is surgical excision.<sup>5</sup>

Surgery should be local resection, keeping margins free of residual disease. Surgical removal can cause extensive wounds, cicatricial strictures, and fecal incontinence. Abdominoperineal amputation is indicated when the sphincter apparatus is involved.<sup>6</sup> Among the most used topical agents are podophyllin, which has an exfoliating, immunological, and antimitotic action,<sup>5,6</sup> and imiquimod, an immunomodulatory substance capable of enhancing the immune response to HPV.<sup>5,6</sup>

### CLINICAL CASE 1

A 25-year-old man with positive serology for HIV for eight months, using antiretroviral therapy (ART) with tenofovir, efavirenz, and lamivudine since his diagnosis, presenting CD4 cell count of 253/ $\mu$ L and undetected viral load.

He complained of an anal tumor for nine months, evolving in the last month with an increase in the lesion, associated with a foul odor and difficulty in hygiene. Upon dermatological examination, he presented a large tumor with a verrucous aspect, 30 cm in diameter, occupying the buttocks, perineum, and intergluteal sulcus, making it impossible to identify the anus (Figure 1).

The biopsy of the lesion ruled out SCC, which report concluded it was a condyloma acuminata. We prescribed sulfamethoxazole + trimethoprim, due to secondary infection, and topical treatment with podophyllin 25% in solid petroleum jelly started once a week, instructing the patient to remove the medication, washing it after six hours (Figure 1).

After 23 applications for six months, we observed almost complete regression of the lesion (Figure 2). After that, surgical resection and cauterization of the few remaining lesions were indicated. The histopathological evaluation of the resected piece did not present carcinoma, and the clinical evolution was favorable. The wounds healed after four weeks, with no signs of recurrence.

### CLINICAL CASE 2

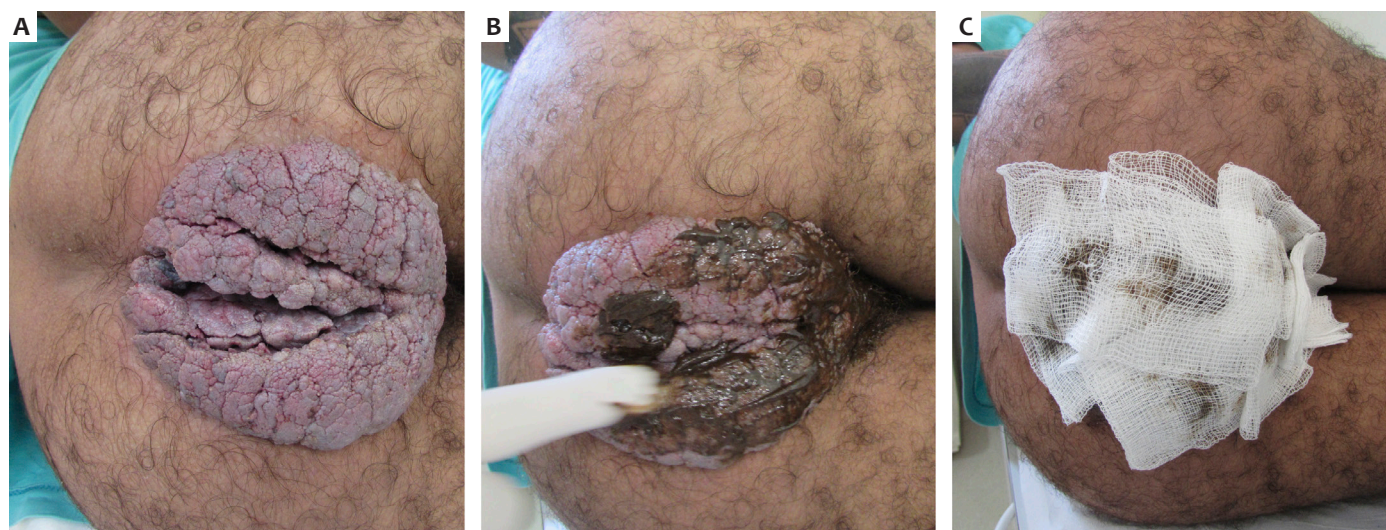
A 42-year-old woman, positive for HIV for six years, complaining of an anal tumor for 12 months. She withdrew ART after six months, with a significant increase in the lesion. The patient had 124/ $\mu$ L CD4 T lymphocytes and a viral load greater than 200,000/mL.

She was instructed to resume ART and started weekly treatment with podophyllin 25% in solid petroleum jelly. After the third application, the lesion showed a significant reduction. She received 12 applications over four months when the disappearance was complete (Figure 3). The biopsy performed at the first consultation revealed condyloma acuminata, with no signs of malignancy. Anal colposcopy and cytology of the anal canal were normal at the end of treatment. There was no recurrence.

### DISCUSSION

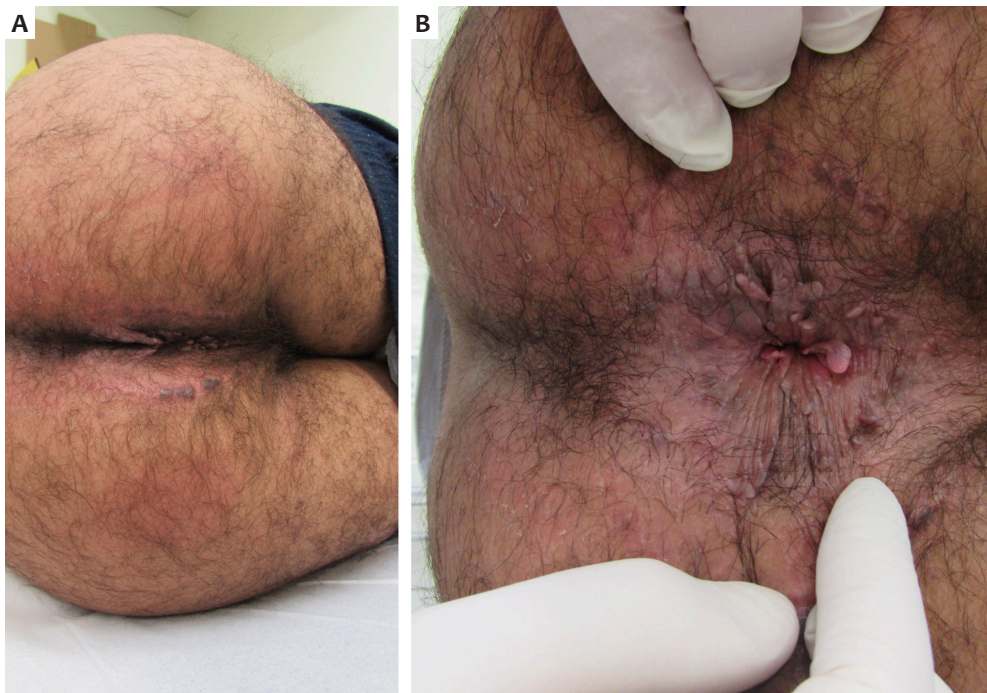
BLT involving the perianal region is rare. A meta-analysis evaluating publications from 1958 to 2000 found only 51 cases. It is more frequent in men (2.7:1) with an average age of 43.9 years.<sup>7</sup>

There appears to be a complex interaction between HIV, HPV, and local mucosa's immunological mechanisms. HIV increases HPV transcription, and it causes a decrease in the number of macrophages, Langerhans cells, and CD4 T lymphocytes in the mucosa, with a consequent reduction in local immune control of HPV infection and increased proliferation of this virus.<sup>8</sup>

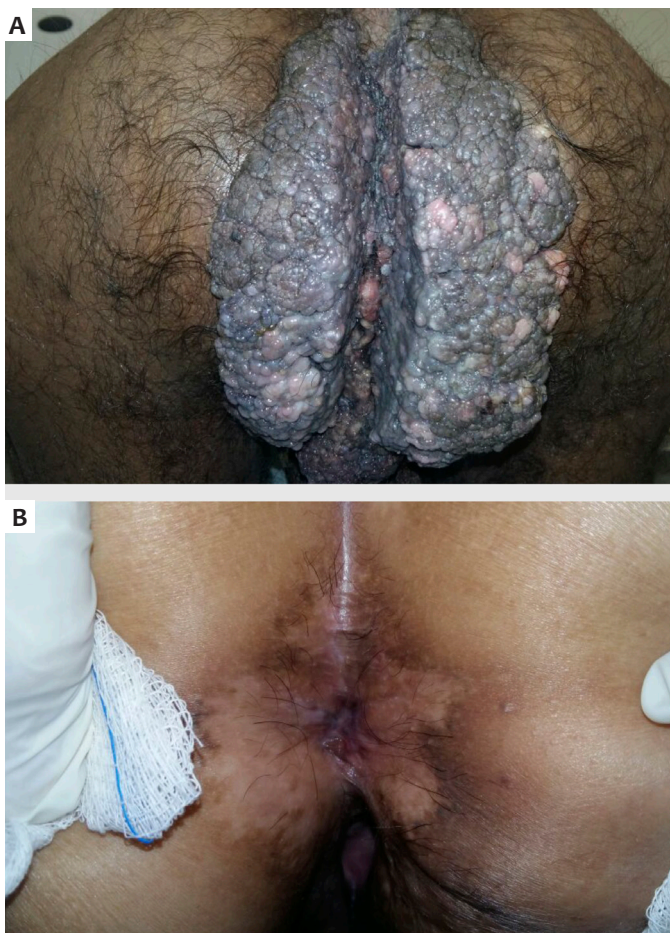


**FIGURE 1:** Buschke-Lowenstein tumor, case 01. (A) Clinical aspect of the lesion. (B) Application of 25% podophyllin in solid petroleum jelly to the lesion and (C) gauze protection before the patient puts on his clothes





**FIGURE 2:** Buschke-Lowenstein tumor, case 01. **(A)** Clinical aspect after 23 weekly sessions performed for six months. **(B)** Detail of the perianal region with spacing of the buttocks



**FIGURE 3:** Buschke-Lowenstein tumor, case 02. **(A)** Clinical aspect of the pre-treatment lesion and **(B)** after 12 weekly sessions performed for four months

Although resection is the most commonly performed procedure, topical treatment was instituted to reduce the lesions' size, thus facilitating resection and postoperative discomfort. Podophyllin 25% in solid petroleum jelly was used because it is easy to obtain and has low cost since the authors work in public hospitals and have experience with this medication in anogenital condyloma acuminata, including in children.<sup>1,5,6</sup> This procedure causes remission of the lesions, avoids surgeries, and facilitates surgical procedures and postoperative recovery.

The patients are being followed up and periodically reassessed with dermatological and proctological examination. If there is no clinical lesion, cytology and anal colposcopy are performed. This follow-up strategy is necessary for surveillance of malignant lesions and early detection of clinical and subclinical recurrences.<sup>7,9</sup>


## CONCLUSION

Treatment with topical substances can be instituted for BLT cases, with the possibility of reducing lesions, facilitating operative treatment, and avoiding complications caused by healing. ●

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