

## Perianal giant condyloma acuminatum (Buschke-Löwenstein tumor). Case report and review of the literature

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### Rezumat

#### **Condilomatoza gigantă perianală (boala Buschke-Löwenstein). Prezentare de caz și date din literatură**

Condilomatoza gigantă perineală, cunoscută ca boala Buschke-Löwenstein (BLT), este o afecțiune rară cu transmitere sexuală, ce afectează regiunea anogenitală. La acest nivel apare o formațiune conopidiformă cu creștere lentă, care spre deosebire de condilomul obișnuit este agresivă și distructivă local, degenerând malign în 40-60% din cazuri. Prezentăm un caz cu BLT malignizată, discuții pe marginea etiopatogeniei și tratamentului acestei boli, cu scopul de a sublinia câteva repere în managementul condilomatozei gigante perineale.

**Concluzii:** Pacientul cu BLT trebuie evaluat clinic-imagistic pentru a stabili gradul invaziei viscerale și astfel amploarea intervenției chirurgicale. Excizia largă a leziunilor, cu margini de siguranță negative histologic este opțiunea de elecție dacă nu este invadat canalul anal. Chirurgia mai radicală este rezervată cazurilor cu invazie viscerală dovedită. Excizia este obligatorie chiar în cazul condiloamelor mici, pentru a preveni evoluția lor către BLT.

**Cuvinte cheie:** condilomatoză gigantă, boala Buschke-Löwenstein, excizie perineală largă

### Abstract

Giant condyloma acuminatum, also known as Buschke-Löwenstein tumor (BLT) is a very rare sexually transmitted disease that affects the ano-genital region. BLT is a slow growing cauliflower-like tumor, but unlike simple condyloma, it is locally aggressive and destructive, malignant transformation occurring in 40-60% of cases. We present a case of perianal carcinomatous BLT and discuss some controversies about the nature and treatment of this disease. Our aim is to emphasize some basic points in the management of giant perianal condyloma acuminatum.

**Conclusions:** The patient with BLT must be very carefully clinical and imagistic investigated in order to detect the tumor visceral invasion and to establish the extension of the surgical procedure. Wide perineal excision with histopathological margins control is the best surgical choice if the anal canal is not involved. The radical pelvic surgery is indicated only in patients with provable visceral invasion. Excision is mandatory even in very small condilomas to prevent BLT later development.

**Key words:** giant condyloma acuminatum, Buschke-Löwenstein tumor, wide perineal excision

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### Introduction

Buschke-Löwenstein tumor (BLT) is a very rare sexually transmitted disease that affects the ano-genital region (1,2,3). The incidence of this tumor is probably 0.1% in the general population and it was first described by Buschke (4,5). BLT, also known as giant condyloma acuminatum, is a slow growing

cauliflower-like tumor, but unlike simple condyloma, it is locally aggressive and destructive (6). In the pathogenesis of this tumor there are involved HPV types 6 and 11 and, possibly, immune defects (2,3,7). Some authors reported BLT in HIV-infected patients (8).

There are controversies regarding the nature of BLT. Some authors have shown that BLT is a verrucous carcinoma and others have described benign forms of this lesion (9,10). Most of the reports have shown that BLT develops on the basis of initially benign condiloma acuminatum and malignant transformation occurs in 40-60% of cases (4). Distant metastases are rare and the risk of developing anal stenosis is low (4,11,12). Common clinical signs that can be found in such patients are bleeding, lymph node enlargement, usually inflammatory, abscess and perianal fistula (4,12). The loco-regional extension must be carefully assessed to establish the therapeutic strategy. In respect to this, abdominal and pelvic computed tomography and magnetic resonance imaging, pelvic inguinal endoscopy and intraanal ultrasound- endoscopy are useful (13).

Besides pathogenesis, there are also controversies on the BLT treatment (5). The majority of authors agree that surgery is the treatment of choice and is efficient in the early stages of the disease (14). Excision must be wide (15,16). Lymph node dissection is indicated only in cases of suspected malignant transformation (4). Some authors prefer abdomino-perineal amputation, but others support the conservative surgery as the best choice, mainly in terms of the patient's quality of life (13). The high local recurrence rate after excision, of about 50-60%, has suggested the need of pre- and postoperative use of radiation and chemotherapy (12,17). There are reports that loco-regional control of recurrent forms was achieved with combined chemoradiotherapy (18,19,20). There are also rare reports of primary radiotherapy producing complete tumor regression (21). Post-treatment monitoring is strongly recommended (4).

We present a case with perianal BLT with malignant transformation and discuss some controversies about the nature and treatment of this disease. Our aim is to emphasize some basic points in the management of giant perianal condyloma acuminatum.

## Case report

We present a 54 year old man with giant condyloma evolving of about 20 years with no previous surgical or oncologic treatment. The patient, without a history of other pathology, is hospitalized in our clinic presenting a giant cauliflower-like tumor located at the posterior and anterior perineum, with right parascrotal extension. There are purulent cankers, bleedings, perianal pain and right inguinal lymphadenopathy. The patient has also two scrotal tegument small tumors (Fig. 1).

Inpatient bleeding was important from many hemorrhage points. There was an early hemorrhagic shock with pallor, increased pulse frequency and arterial hypotension, malaise and marked fatigue. Laboratory analysis revealed a severe anemia and mild leukocytosis. The patient is admitted to the intensive care unit being hydroelectrolitic rebalanced,



**Figure 1.** Giant perianal and parascrotal condyloma

transfused and receiving antibiotics. A biopsy was performed and histopathology reveals a malignant transformation. The Human Immunodeficiency Virus test result was negative.

Surgical intervention was then performed involving, in a first stage, the perianal tumor resection using the electrocision. The right parascrotal condyloma was excised after 12 days, in the second stage.

The pathological evaluation showed a solid, exophytic and invasive tumor, hard and white on the surface of section. The microscopic diagnosis was squamous cell carcinoma moderately differentiated (Fig. 2). Tumor cells showed anaplastic features and many mitoses (Fig. 3). Cytokeratin AE1/AE3 was strong but heterogeneously expressed in the tumor area (Fig. 4). Both p53 and Ki67 were expressed by the majority of tumor cells, reflecting the high proliferative potential of the tumor (Fig. 5, 6). No lymphovascular invasion was detected on slides stained with anti-D2-40/podoplanin, but positive reaction was noticed in less-differentiated neoplastic cells, supporting the aggressive and invasive behavior (Fig. 7).

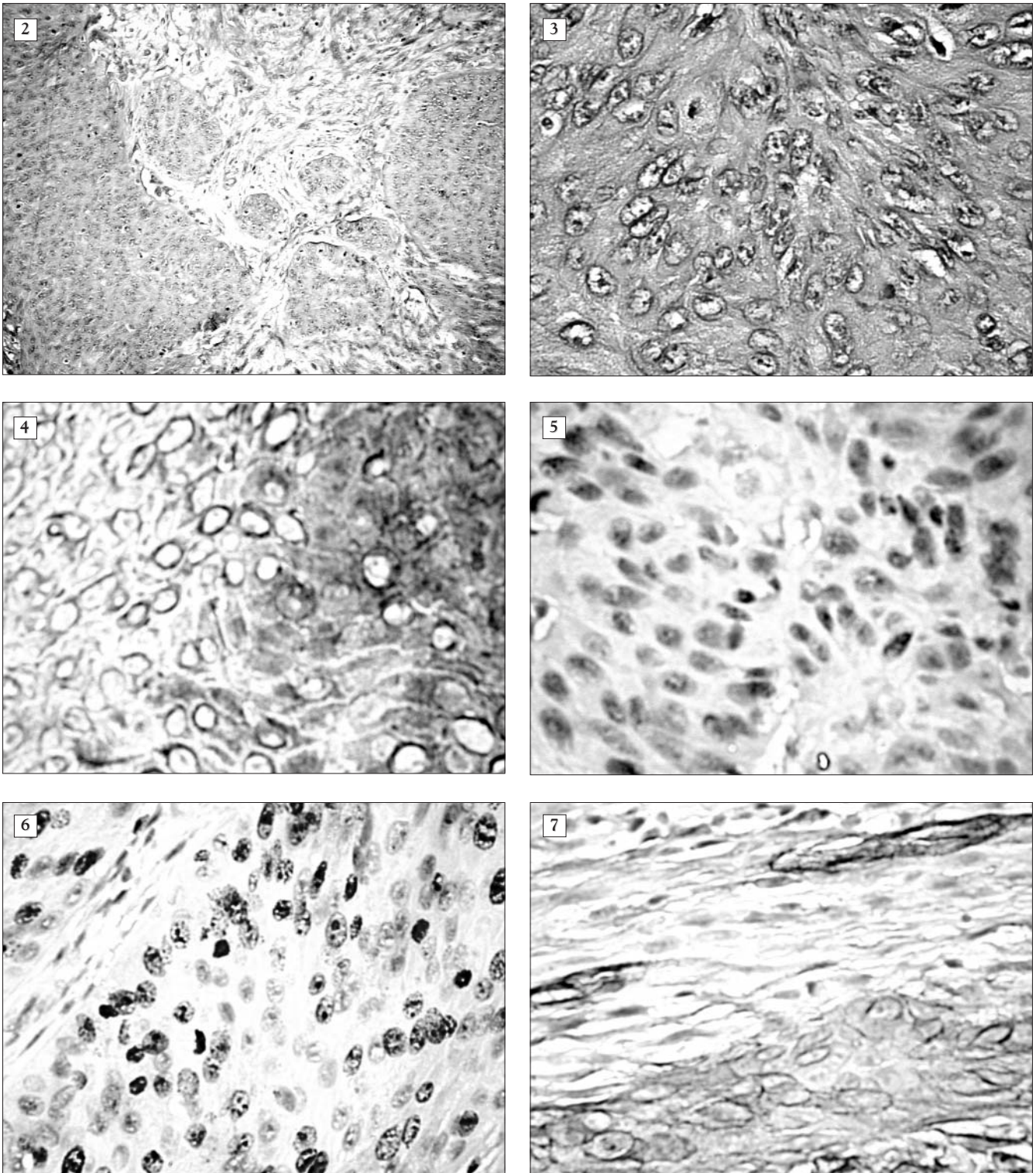
The postoperative evolution was favorable with granulation tissue and small necrotic areas that require serial necrectomies (Fig. 8).

Controls at 1 and 3 months revealed a slow favorable evolution with epithelization of the wound and remission of the inguinal adenopathy. After 6 months the perineal region is free of tumor recurrence. The patient refused chemoradiotherapy.

## Discussions

The case presented is a less cooperant, homeless patient with a very long disease evolution, who was hospitalized forced by local massive bleeding. The giant tumor evolved from small cauliflower-like vegetations, as we know that BLT is always preceded by condyloma acuminatum (4,22).

The anal canal was carefully examined and it was not involved by the tumoral invasion (5). Malignant transformation was present, being suspicioned by massive bleeding,



**Figures 2-7.** 2,3 – low power magnification of the tumor - squamous cell carcinoma (x100); 4 - heterogeneous expression of cytokeratin AE1/AE3; 5 - p53 expression; 6 - Ki67 expression ; 7 - anti-D2-40/podoplanin stained both lymphatic vessels and tumor cells. Original magnification 7-10 x 400

tumor infiltration, its giant size and lymph node infiltration (4,12). The cancerous nature of the tumor was confirmed by histopathological exam that reveals a squamous cell carcinoma, after a careful sectioning and complete histologic exami-

nation was performed, as it is always required in such cases (9, 23). This tumor shows aggressive local infiltration but does not metastasize (23).

There is a controversy in the histopathological diagnosis



**Figure 8.** Aspect 2 weeks after the second excision with granulation tissue and superficial necrotic areas

of BLT. Most of the authors sustain that BLT is a pseudo-épithéliomatous proliferation belonging to verrucous carcinoma group (24), and we have the same opinion, supported by the findings in our case. But there are some reports regarding BLT as an intermediate entity between an ordinary condyloma acuminatum and squamous cell carcinoma (18). Finally, there is a case report of microscopically benign giant condyloma acuminatum of the perianal region (10). Anyway, the histopathological dilemma is not so important, because wide excision is always mandatory when is possible.

So far, no definitive therapeutic strategy has been established (5). The management of BLT must be implemented knowing the pelvic tumor extension which needs clinical and imaging exams. Unfortunately, we did not have quick access to computed tomography or magnetic resonance facilities and the developing haemorrhage made the operation mandatory.

The majority of authors agree that surgery is the treatment of choice and is effective especially in the early stages of the disease (15). Wide local excision remains the mainstay of therapy, that can be followed, if it is necessary, by delayed split thickness skin grafts (1,11,25,9). There are some controversies between radical abdomino-perineal amputation and conservative surgery supporters (13).

We think that wide perineal excision with histopathological margins control is the best surgical choice in the treatment of BLT if the anal canal is not involved. The radical pelvic surgery is reserved only for patients with provable visceral invasion. Wide resection is indicated even in rare cases of benign nature BLT because of the high risk of carcinomatous transformation and the great discomfort produced by the giant perineal tumor. We also emphasize that surgical excision is mandatory in all cases of BLT when it is technically possible and also in all condylomas, even in very small ones, in order to prevent their BLT transformation.

There is no consensus about the use of pre- and post-operative oncologic treatment in BLT. Some authors recommend that reduction of the tumoral mass through

radiotherapy or chemotherapy to precede surgical excision (4,15). Others assert that radiotherapy is rarely used, usually after an incomplete excision, in recurrences and when excision is not recommended. There are rare reports of primary radiotherapy producing complete tumor regression (21). Radiotherapy has been suspected of being responsible for the alteration of BLT into anaplastic carcinoma (15,16,26). We can not express a clear opinion about the oncological treatment in our case because our patient refused radio-chemotherapy.

Some authors reports the use of imiquimod and others present a case of BLT successfully treated using carbon dioxide laser vaporization and systemic interferon therapy after failure of 3 surgical excisions (2,27,28,29,30).

The patient must be long time closely followed-up (4, 31). We think prospective studies are necessary to further define the nature and treatment of this very rare disease (32).

## Conclusions

BLT is a very rare sexually transmitted disease characterized by giant slow growing condyloma acuminatum that is, unlike simple condyloma, locally aggressive and destructive.

The histopathological nature of BLT is not well defined, but it is clear that malignant transformation rate is high.

The patient with BLT must be very carefully clinical and imagistic investigated in order to detect the tumor visceral invasion and to establish the extension of the surgical procedure.

Wide perineal excision with histopathological margins control is the best surgical choice if the anal canal is not involved. The radical pelvic surgery is indicated only in patients with provable visceral invasion.

Excision is mandatory even in very small condilomas to prevent BLT later development.

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