

Buschke-Löwenstein tumor in a human immunodeficiency virus-positive patient : a case report and short literature review

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Abstract

Giant condyloma acuminatum, also known as Buschke-Löwenstein tumor, is a rare variant of verrucous carcinoma presenting in the ano-genital region. While its metastatic potential is limited, aggressive local growth is common, with invasion and destruction of the surrounding tissues often causing important therapeutic challenges. Also, data to inform the optimal management approach are scarce and mostly limited to anecdotal reports. We present the case of a human immunodeficiency virus-associated locally advanced Buschke Löwenstein tumor that was successfully treated with extensive surgery. (Acta gastroenterol. belg., 2021, 84, 343-345).

Keywords : Buschke-Löwenstein tumor, giant condyloma acuminatum, human papillomavirus, squamous cell carcinoma, human immunodeficiency virus.

Introduction

Giant condyloma acuminatum (GCA) or Buschke-Löwenstein tumor (BLT) was described for the first time in 1925 as a rare variant of verrucous carcinoma presenting in the ano-genital region (1). The incidence of this tumor in the general population is estimated to be about 0,1%, and is most common among young adults, with 2.3 :1 male to female ratio (2,3).

Human papilloma virus (HPV) infection (especially from low-risk genotypes HPV-6 and HPV-11) and genital warts (GW) are the most important etiopathogenic factors (4). Additional risk factors include smoking, multiple sexual partners, anaerobic infections, local chronic inflammation and immune deficiency (5).

Generally, this tumor arises from the external genitalia, while in only 10-17% of cases it is localized in the anorectal area (6). Clinical hallmarks include a locally invasive and destructive growth, frequent local recurrence after treatment, and low metastatic potential (2). If left untreated, BLTs can extend into the pelvis, causing disabling symptoms and complications such as fistulisation and abscesses. Some tumors can also transform into more aggressive squamous cell carcinoma (SCC) (7).

The treatment of choice is surgery, other approaches such as radiotherapy and chemotherapy having modest efficacy (6).

Here, we present the challenging case of a locally advanced BLT occurring in a human immunodeficiency virus (HIV) - positive patient.

Case report



Figure 1. — (A) perianal condyloma with large abscess (before surgery). (B) perianal view 3 months after abdominoperineal resection. (C) Sagittal T2-weighted MRI showing the lesion involving the pelvic floor. (D) Transversal T2-weighted MRI showing the lesion.

A 51-year-old man presented to the local gastroenterology outpatient clinic, complaining of nocturnal anal pain and dysuria for a few days. His medical history included HIV infection since 2011, resection of GWs, anal intra-epithelial neoplasia (AIN) II in 2013 and AIN I in 2014, hepatitis C infection in 2015 treated successfully with Sofosbuvir and Daclatasvir, syphilis infection and proctitis from chlamydia trachomatis and neisseria gonorrhoea. He was an active smoker, snorting cocaine on occasion. Regular medications included lormetazepam, raltégravir and emtricitabine/tenofovir.

Physical examination revealed a large, exophytic, cauliflower-like perianal verrucous tumor extending to

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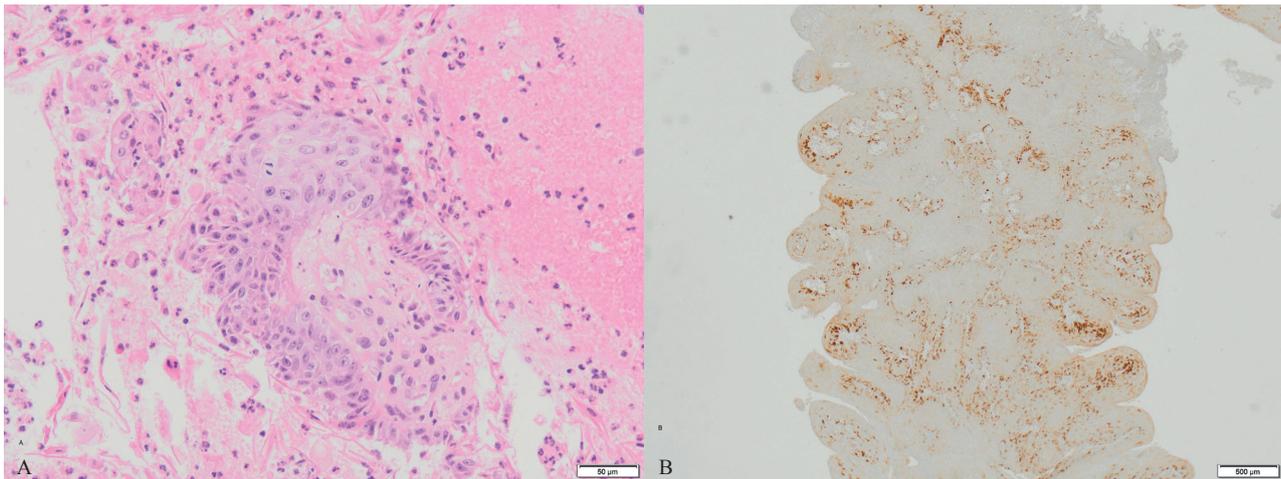


Figure 2. — (A) A papillary structure showing in the basal and the medium layer nuclear atypia and pleomorphism. There are mitosis in the medium layer. (original magnification x 200) (B) The scoring of p16 generally includes both nuclear and cytoplasmic staining, and is graded as 1 (rare singly dispersed cells staining). (original magnification x 20)

the right cheek with induration and erythema suggestive of an abscess (Figure 1).

A blood test showed microcytic anemia (11,3 g/dl), thrombocytosis ($539 \times 10^3/\mu\text{L}$), leukocytosis (15,810/ μL , neutrophils 8,440 cells/ μL) and high C-reactive-proteine (98.3 mg/L). HIV viral load was undetectable, while CD4 count was 500/ μL .

An abdomino-pelvic CT scan suggested sigmoido-rectitis with right para-rectal and anal abscesses. An MRI of the pelvis revealed a large tumor in the right para-rectal region (7 cm x 8 cm x 14 cm) with diffuse infiltration of the rectal wall and extension into the subcutaneous tissues, and a small (1 cm) right inguinal adenopathy (Figure 1C, 1D). An ^{18}F -FDG PET-CT scan was also carried out, which confirmed a large hypermetabolic mass (SUV max 24) in the right para-rectal region and along the anal canal, as well as moderately hypermetabolic inguinal and external iliac lymphadenopathies. Finally, an endorectal ultrasound showed an extra-sphincteric fistula with an abscess in the prostate bed.

Initial unsuccessful management included antibiotic therapy, repeat surgical incisions and drainage of the abscesses. Further to the persistence of symptoms including weight loss, severe pain and constipation, a discharge colostomy was performed, and the patient was finally referred to our center.

Following a preliminary assessment in the surgical outpatient clinic, the patient case was discussed at our multidisciplinary team (MDT) meeting, where biopsy of the tumor mass was recommended. A number of biopsy attempts were made, and histology was eventually consistent with a p16-positive condyloma and high-grade dysplasia, without evidence of invasive carcinoma (Figures 2A ; 2B (8)). Given the high suspicion of BLT, however, the ultimate MDT decision was to proceed with surgical resection.

An abdominoperineal resection with cystectomy, orthotopic neobladder and vertical rectus abdominis

myocutaneous (VRAM) flap reconstruction was performed. In the immediate postoperative period, the patient suffered from hemorrhagic shock due to necrosis of the graft. A repeat surgery was carried out to control the bleeding, remove the graft and perform an omentoplasty.

Pathological examination of the surgical specimen confirmed the diagnosis of BLT of the anal margin infiltrating the rectum and making contact with prostate and bladder (stage pT4NxMx). Surgical margins were clear, and there was no evidence of either vascular or perineural invasion. One year after surgery, the patient is alive and free of disease. (Figure 1B)

Discussion

In the literature, little information is available regarding the optimal management of BLT. Most of the available evidence is based on case reports and small retrospective series. Nevertheless, the treatment of choice remains surgery with clear margins (6), with the risk of recurrence being as high as 66 %, and an overall mortality rate of about 20% (9). Also, it is generally accepted that early surgical resection of GWs can reduce the risk of BLT development (10).

Other treatments have been described, with the management approach being often influenced by factors such as size and location of the tumor, and prior unsuccessful therapies (11). Chemotherapy alone (generally cisplatin and 5-fluorouracil) or concurrently with radiotherapy has been shown to induce tumor response in some cases (6). Chemoradiotherapy has been proposed to downsize the tumor in patients with malignant transformation of BLT(2). Intra-arterial chemotherapy with agents such as methotrexate has been successfully used in verrucous carcinomas, including those from the ano-genital region (12). Finally, there are some reports suggesting the activity of intralesional or systemic interferon (13), while oral, topical and intra-

lesional chemotherapy agents have been used with variable success as adjuvant treatments after surgery or for the management of recurrent tumors (7).

Our case highlights the challenges that physicians may encounter during the diagnostic work-up of BLT as well as the importance of an MDT-driven approach. Despite multiple unsuccessful biopsy attempts, a diagnosis of BLT was suspected based on patient risk factors (smoking, multiple sexual partners, HIV infection), the presence of GWs, and the local invasiveness of the tumor. While upfront surgery implied an extended and mutilating resection to achieve clear margins, it was considered as the most appropriate treatment to promptly relieve local symptoms and to offer the best chances in terms of oncological outcomes. Other options, such as chemoradiotherapy or chemotherapy alone (either before or instead of surgery) were not taking into account in view of the large size of the lesion, multiple abscesses, and severe symptoms.

Conclusion

BLTs are rare tumors posing significant diagnostic and therapeutic challenges. While surgery with clear margins remains the standard treatment, an MDT-driven management approach should always be adopted.

Conflict of interest

No conflicts of interest regarding the publication of this paper.

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