CASE REPORT

Successful improvement of Buschke-Löwenstein tumour in an HIV-infected patient with antiretroviral therapy alone

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SUMMARY

Buschke-Löwenstein tumour (BLT), also defined as giant condyloma acuminatum, is a rare exophytic tumour affecting the anogenital and perianal regions associated with human papillomavirus (HPV) infections, with a potential of malignant transformation and which is at a greater risk in T-cell mediated immunodeficient patients. Different therapeutic options, alone or in combination, have been reported for the treatment of BLT including local therapy but wide surgical local excision is however recommended as the most important therapeutic intervention. We report a case of a HIV-infected man who developed a voluminous pelvic BLT which disappeared progressively under antiretroviral therapy with no additional treatment, contemporary to an improvement of his immunity, highlighting the possible spontaneous reversibility of HPV-induced tumours in treated HIV infection.

BACKGROUND

Since wide surgical local excision is the most recommended therapeutic intervention for the treatment of Buschke-Löwenstein tumour (BLT) to prevent disease recurrence and because of its potential of malignant transformation, treatment of such large tumours may lead to severe mutilations. In addition as cell-mediated immune response plays a major role in the natural control of human papillomavirus (HPV) infection, correction of immunodeficiency in HIV-infected patients with BLT by antiretroviral therapy appears to be the primary treatment in this context. We report this case to show that regression of BLT was conceivable with antiretroviral therapy alone, which has not been reported earlier to the best of our knowledge.

CASE PRESENTATION

A 47-year-old man from Guinea-Bissau with HIV, hepatitis B and D coinfection was admitted in our clinical department in September 2014 for multiple condylomas located on the scrotum, perineum and perianal regions. HIV-1 infection had been diagnosed in 2008, initially complicated with pulmonary tuberculosis and pneumocystosis. Warts appeared 6 months ago, and gradually worsened until having an aspect of giant verrucous condylomatous lesions compatible with a BLT (figure 1). No enlarged inguinal lymph nodes were noted.

Owing to insufficient adherence to antiretroviral therapy and interruption of medical follow-up since several months, the patient also developed an

end-stage renal disease related to HIV-associated nephropathy (HIVAN) leading to recurrent haemodialysis. CD4+ lymphocyte count at admission was 26 cells/ μ L (5%) and HIV RNA level was 5.21 log copies/mL.

INVESTIGATIONS

The diagnosis of BLT was histologically confirmed with condylomas' biopsies consistent with verrucosis and papillomatous hyperkeratotic lesions with many superficial koilocytes, a slightly congestive chorion punctuated of some mononucleated inflammatory elements and a few basal mitosis. No cellular atypia was found.

The patient underwent endoscopic procedures for tumour extension evaluation including anoscopic and colonoscopic examinations revealing a small anterior condyloma in the distal anal canal (completely withdrawn) as well as a cystoscopy showing no vesical condyloma. MRI of the pelvis excluded a deep extension of the disease.

TREATMENT

A highly active antiretroviral therapy including darunavir, ritonavir, tenofovir disoproxil fumarate and lamivudine was rapidly initiated, associated to a reinforcement of the adherence to antiretroviral therapy. T CD4 cell count progressively increased up to 229 cells/mm³ (20%) over 6 months, and HIV RNA level decreased to <20 copies/mL and remained undetectable.

OUTCOME AND FOLLOW-UP

While surgical excision of the tumour was initially planned it was postponed as a significant decline of the condylomas was observed under antiretroviral therapy, contemporary with the HIV-1 suppression and the CD4 immune recovery. By December 2015, an almost complete tumour regression was recorded, with only few remaining condylomas located on the scrotum and on the pubic area (figure 2). Topical imiquimod was not prescribed due to application difficulties. As no systemic or local treatments were given, we concluded that cellular immune recovery due to the antiretroviral therapy alone was responsible for the BLT regression.

DISCUSSION

Development of BLT, also defined as giant condyloma acuminatum, is usually associated with HPV types 6 and 11 infection, while HPV types 16–18

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Figure 1 Exophytic, cauliflower-like friable mass consistent with a Buschke-Löwenstein tumour involving the intergluteal cleft, the left groin, the perianal region and the scrotum before starting antiretroviral therapy (September 2014).

Figure 2 Few remaining slight condylomas located on the left groin, the perianal region and the scrotum after initiating antiretroviral therapy (December 2015).

can be sometimes involved in the generation of this type of lesion. DNA of these viruses is identified in giant condylomas.¹ In immunocompetent patients efficient cell-mediated immune responses prevent HPV infections. In T-cell mediated immunodeficient patients, such as HIV infected patients, there is a high risk of developing HPV-associated lesions. BLT may be associated with severe morbidity resulting from aggressive local tissue invasion and destruction, occlusion of the rectum, and malignant transformation into squamous cell carcinoma.²

Different therapeutic options, alone or in combination, have been reported for the treatment of BLT, such as surgery, radiation therapy, topical and intralesional chemotherapy, systemic interferon α -2b or interleukin 2, topical therapies including podophyllin, fluorouracil, imiquimod, cidofovir, carbon dioxide laser therapy, and photodynamic therapy.^{3 4} A wide surgical local excision is however recommended as the most important therapeutic intervention.

No surgical treatment was finally required in our patient, as antiretroviral therapy alone led to a regression of the lesions. Spontaneous regression of genital warts occurred, enabled by immune cellular infiltration and activation.⁵ However,

conversely, some cases of paradoxical growth of the tumour under antiretroviral therapy have also been reported, resulting from the development of an immune restoration syndrome due to an inflammatory reaction to HPV infections.⁶

Indeed in our case, the improvement of cellular immunity due to antiretroviral therapy enabled us to successfully treat the BLT, highlighting the role of immunodeficiency in viral-induced tumours' occurrence in HIV clinical settings.

Learning points

- ▶ Buschke-Löwenstein tumour is a rare human papillomavirus (HPV)-related tumour with potential malignant behaviour.
- ▶ Immunocompromised patients, including HIV-infected patients, are at a higher risk of Buschke-Löwenstein tumour.
- ▶ Despite different therapeutic options, wide surgical local excision is the most recommended therapeutic intervention.
- ▶ However, immune recovery under combined antiretroviral therapy is able to reverse alone Buschke-Löwenstein HPV-induced tumours in HIV settings.

Contributors CG, AH, J-DL and SG were in charge of the care of the patient and were responsible for the acquisition of data. CG and SG were responsible for the study concept and design and drafted the manuscript. CG, AH, J-DL and SG performed critical revisions of the manuscript for important intellectual content. SG was responsible for study supervision.

Competing interests None declared.

Patient consent Obtained.

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