



Inflammation and Infection

Giant Condyloma Acuminatum in the Genital, Perineal and Perianal Region in a Pediatric Patient. Literature Review and Case Report



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ABSTRACT

Condyloma acuminata is caused by the proliferation of squamous epithelial cells in the presence of human papilloma virus (HPV) infection. There are several treatment options available for anogenital warts, however, none have proven to be more efficacious. We present the case of a 3 year-8 months-old male, diagnosed with human immunodeficiency virus (HIV) infection, who presented with multiple warts in the anogenital region. Lesions were treated with imiquimod 5%, electrosurgical resection and interferon α -2b. Combination of electrofulguration and interferon α -2b is an effective treatment option for children with giant condyloma acuminatum although recurrence is expected within a short follow-up period.

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Introduction

The Buschke-Löwenstein tumor (BLT), or giant condyloma acuminatum, is a semi-malignant verrucous neoplasia characterized by aggressive growth into underlying dermal structures. It is recurrent in 30–70% of cases and can progress slowly into exophytic, ulcerative and cauliflower-shaped tumors that can form abscesses and fistulas.^{1,2} It is generally located on the genital area, however, on occasions it involves distinct histological zones of the anorectal region.¹ Now a days there is no approved treatment by the *Food and Drug Administration* in the pediatric population, nevertheless a number of treatment options are available for anogenital warts.³ In 2010 the *Center for Disease Control and Prevention* published its guidelines on sexually transmitted diseases and recommended that treatment be directed based on size, location and number of lesions. The therapeutic options include topical ointments, cryotherapy, laser vaporization, electrosurgery and surgical excision. The aim of

treatment is to eliminate visible warts, nonetheless that does not guarantee eradication of HPV infection.³

Case report

A 3 year-8 months-old male presented to the emergency department with a large cauliflower-like growth covering the penis, scrotum, perineal and perianal region. The patient was tested HIV ELISA positive as a consequence of vertical transmission from his 20-year-old mother, also HIV positive. The mother's HPV infection was never documented since she abandoned the patient after their first hospital visit. No history or evidence of sexual abuse of the child was encountered. A final diagnosis of pediatric HIV presenting with giant genital and perianal condyloma acuminata was made. He was initiated on broad-spectrum antibiotics and antiretroviral therapy with lopinavir, ritonavir, abacavir and lamivudin.

The growth had started 2 years prior as warty lesions around the genital area, and had increased in size and coalesced to assume large dimensions invading the scrotum, base of the penis, prepuce, perineal area and anal sphincter. At presentation to the emergency department in January 2015, the HIV viral load was undetectable, and a CD4 percentage of 23% (range: 31–60%) was reported (Fig. 1).

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Figure 1. Initial presentation.



Figure 2. Postoperative picture after fourth surgery (3 months after treatment).

Initially, treatment was started with imiquimod 5% ointment three times per week without any visible improvement. Therefore, electrosurgical resection was performed under general anesthesia over multiple sessions due to the considerably large size of the condyloma. Complete resection was achieved after seven interventions separated by 2-week intervals. The specimens were sent for histopathological evaluations, and condyloma acuminata with neovascular proliferation was reported. No serotype assessment was made due to test unavailability at our institution. After completion of electrosurgical treatment, immunotherapy was initiated with IFN α 2b at a dosage of 2 million units per week during 4 weeks (Figs. 2 and 3).

Monthly outpatient follow-up was scheduled for a year. Recurrence in the genital area was documented 3 months following complete resection of the lesion. Cutaneous examination of the perianal region showed no remaining scrotal skin per-se, instead, the area showed reddish-brown plaques with soft non-fibrotic tissue. No further alterations were observed on the penis, anus or urethra.

An additional 0.5 cm recurrence in the perianal zone was noticed on the 12th month of consultation, and ophthalmologists were requested to assess a solitary papillary lesion on the caruncle and tarsal conjunctiva of the right eye, originally treated with an unsuccessful cycle of IFN α 2b. The patient is now awaiting surgery for both lesions.

Discussion

The Buschke-Löwenstein tumor is a rare entity with an average incidence of 0.1% in the general population.⁴ Infection by HPV has been linked to the development of BLT and recently 70 subtypes

have been reported.¹ The HPV is classified as high and low-grade, depending on the virus oncogenic potential. The low-grade subtypes include 6, 11, 42, 43 and 44; and are isolated from 90% of genital warts.³ The high-grade subtypes 16, 18, 31, 33, 35 and 45 are capable of causing high-grade dysplasia and squamous cell carcinoma. The presence of co-infection by high-grade HPV in genital warts is 31%.³ The immune system effectively repels the majority of HPV infections by localized cell-mediated immune responses. Approximately, 10% of individuals will develop a persistent infection with a risk of proliferative lesions, high-grade precursors and eventually an invasive carcinoma. In our reported case, it was not possible to determine the subtype of the virus.

In the pediatric population the average age to develop anorectal warts has been reported to be between 2.8 and 5.6 years of age.⁵ The proposed mechanism for perinatal and postnatal transmission includes vertical transmission, autoinoculation, sexual transmission and indirectly through contaminated objects and surfaces. The incubation period varies between 3 weeks and 8 months and the majority of anogenital warts appear 2–3 months after exposure. The rates of vertical transmission of HPV to the neonate in women without clinical evidence of infection varies between 1–18%, compared to 5–72% in women with a detectable HPV infection during pregnancy.⁵ Co-infection with HPV occurs in 8–10% of HIV positive pediatric patients.

Treatment is difficult due to the high recurrence rates (30–70%) and risk of bleeding.¹ There are a number of medical and surgical treatment options available to treat anogenital condyloma acuminata.

Surgical management consists of excision, fulguration or both. Without a current gold-standard, surgery remains the cornerstone of treatment. The case reports that have been published show



Figure 3. Last follow-up picture after eighth surgery (8 months after treatment).

satisfactory results with different therapeutic approaches either with mono or combined therapy. However, they are not mutually comparable, and neither has any superior efficacy over the other that can be sustained.

The case presented is of particular interest due to the extent of the lesions and their rapid appearance, associated with a low HIV load in response to the administration of antiretrovirals. Also of interest is the refractoriness shown by the condylomas to topical therapy with imiquimod, not only did they not diminish but rather

increased in size and number. These observations suggest a possible synergistic adverse effect of HIV for the control of human papilloma virus infection. However, the subsequent handling of a combined scheme with IFN and electrosurgical resection was successful for the complete elimination of the condyloma acuminata. Although the behavior of HPV remained unchanged, a rapid recurrence of lesions merited new surgical events. This unusual behavior, even considering the rate of recurrence high in the general population, is likely due to the combination of viral diseases. This should be kept in mind in the regular and frequent clinical monitoring of the patient despite having a good control of viral replication expressed by a low HIV load.

Conclusion

The Buschke-Löwenstein tumor or giant condyloma acuminatum is an extremely rare clinical type of genital wart in the pediatric population, with a high recurrence rate which may be higher due to the synergistic effect of HIV on HPV's aggressive nature. HIV infection should thus be considered an additional risk factor for a poor response to medical treatment only, with a high risk of recurrence. Electrosurgical resection and IFN therapy are efficacious procedures to control the rapid extension of the disease.

Conflict of interest

None of the authors have any conflict of interest to report.

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