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Gaint Condylomata Acuminate of Buschke-Lowenstein in Woman with HPV Infection: Clinical Presentation and Treatment

Abstract

Buschke-Lowenstein Tumor (BLT) is a very rare sexually- transmitted disease associated with human papillomavirus type 6 and 11, but rare cases of oncogenic HPV types including HPV 16 and HPV 18 were also reported. BLT is located in the genital, anorectal and perianal regions. It is regarded as a type of verrucous carcinoma occurring on anogenital mucosal surfaces where it is locally invasive but displays a benign cytology. Buschke-Lowenstein tumor can be associated with a high rate of recurrence and a risk of malignant transformation to invasive SCC, especially in patients with oncogenic types of HPV. We report the case of a 59-year-old female patient who addressed our clinic for a large, exophytic, cauliflower-like tumor involving the vulva, perineum and perianal regions with 20 years duration. The first lesions had been appeared on vulva and after 3 years period they grown slowly and covered perineum and perianal area. They had cauliflower like surface, with different sizes. In some points erosions and yellowish secretion with odor are observed. Histologic examination of a biopsy specimen of large tumor presented nets of well- differentiated squamous cell carcinoma, as well as a marked mononuclear cell infiltrate and conspicuous koilocytosis. HPV DNA for 6 and 11 types was detected with PCR. The patient was sent to the gynecology surgery department for excision and remains under the supervision of the dermatology and oncology department for rapid treatment of relapses and early detection of malignant transformation.

Keywords: Buschke-Lowenstein tumor; HPV; SCC

N Tsekova Traykovich*, D Nikolova, A Argirov*

Department of Dermatovenereology, Medical University, Sofia, Bulgaria

*Corresponding author: N Tsekova Traykovich

natasa_cekova@yahoo.com

Tel: 359 886307415

Department of Dermatovenereology, Medical University, Sofia, Bulgaria

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Introduction

Buschke-Lowenstein Tumor (BLT) is a very rare sexuallytransmitted disease associated with human papillomavirus. It is a neoplasm of the anogenital region which has appearance on histopathology as relatively low-grade malignancy, but is locally destructive and with aggressive behaviour [1]. We have discussed an overview of etiopathogenesis, clinical presentation, diagnosis and management of this uncommonly disease [2].

Case Presentation

A 59-year-old female multiparous, immune competent patient with two normal deliveries and in menopause for 15 years presented to our clinic with a large, exophytic, cauliflower-like tumor involving the vulva, perineum and perianal regions.

The first lesions had been appeared on labia majora as pin-size papules before 30 years ago during pregnancy period [3]. They gradually increased in size and coalesced to form large tufts and covered perineum and perianal area.

It was not painful, but caused some discomfort mainly during sitting, urination and defecation. The lesions were treated

with different locally destructive modalities without significant results. Her husband was examined and found to be free of any lesions in genital area [4].

The patient denied any history of extramarital sexual contact or assault. Physical examination reveled a 30 cm exophytic cauliflower like tumor affecting vulva, perineum and perianal regions and low vagina [5].

In some points erosions and yellowish secretion with odor are observed. Upper vagina and cervix appeared normal. No locoregional lymph nodes were present upon palpation. A Papanicolaou cervical smear was performed and smear from portio vaginals cervix uteri and cervix coli was classified as IIA group [6].

Her HIV, HbsAgan, VDRL status were nonreactive. Contrast enhanced computer tomography of chest, abdomen and pelvis was done that did not reveal any regional lymph node involvement or evidence of distant metastasis. Colonoscopy data for tumor mass which reached linea dentate mass without invasion in anal sphincter (Figure 1).

A biopsy, under local anesthesia, was performed. On histology we observed koilocytic changes, hyperkeratosis and parakeratosis.

The epithelium extends into the dermis with blunt-shaped masses, forming sinuses and keratin-filled cysts. The distinction from conventional squamous cell carcinoma witch rests on 2 criteria: cytologic atypia and clearly infiltrative pattern of growth, are not seen in our case [7]. A PCR of the specimen revealed the presence of HPV- type 6,11. The patient was send to Gynecology department and radical vulvectomy was performed. Follow-up after 1 year did not reveal any new lesions (Figure 2).

Discussion and Conclusion

BLT first described by Buschke and Lowenstein in 1925. They observed a penile lesion that clinically resembled both common condyloma acuminata and squamous cell carcinoma, but differing from both of them regarding the biological behavior and the histopathological appearance. Some authors support the hypothesis that BLT is an intermediary lesion between condyloma acuminata and SCC. However, others equate it to verrucous carcinoma of the anogenital region. It is a very rare sexually transmitted disease associated with HPV type 6 and 11, suggesting a pathogenic role. Rare cases of oncogenic HPV types-HPV 16, 18, 54 were also reported. There is no racial predilection and male-to-female ratio is 3.5:1. BLT is located in the genital, anorectal and perianal regions. It is well differentiated, benign lesion and its management is often challenging due to the size, rapidly growing, degree of local invasion, and recurrence rate. Perirectal GCBL include perianal mass (47%), fistula or abscess (32%), and bleeding (18%). BLT is associated with a high rate of recurrence (up to 70%) and a risk of malignant transformation to SCC (30-56%), especially in immune-compromised status and patients with oncogenic types of HPV.

The diagnosis is based mainly of clinical presentation, histological picture and PCR- HPV DNA. A proper biopsy must be wide and deep enough to accurately determine the extension of infiltration of the



Figure 1 Kilocytes, hyperkeratosis and parakeratosis.



Figure 2 Removed tumor mass.

tumor and the possible presence of a squamous cell carcinoma. differentiated from ordinary condylomas by the characteristic pushing rather than "infiltrating" effect that tends to compress and displace the underlying tissue. Radical surgical excision is the gold standard in treatment. Other various treatment topical modalities include podophyllin, 5-FU, Interferon, Bleomycin (intralesional), Cidofovir gel 1.5%, Imiquimod in association with CO2 laser. Chemoradiation has been recommended when malignant transformation my occur. Radiation therapy alone has been discouraged because of protentional risk of transformation into anaplastic carcinoma. And last but not the least HPV vaccine could lead to a protentional prevention and reduction in the incidence of this disease. Because of frequent recurrence and malignant transformation multidisciplinary patient control collaboration is necessary and regular follow-up is mandatory.

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