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Case report

GIANT CONDYLOMA ACUMINATUM OF BUSCHKE-LOWENSTEIN: A CASE REPORT

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Abstract

Buschke-Löwenstein tumor is a rare, slow-growing tumor that may arise from a pre-existing condylomatous lesion and has exophytic, cauliflower-like, or papillomatous growth pattern.

These tumors are associated with low-risk human papillomavirus infection, most commonly types 6 or 11. They predominantly occur in men in the anogenital region, and are associated with immunodeficiency. The treatment is controversial, but the greatest success has been observed with "cold knife" surgical resection with clear margins.

We present the case of a 42-year-old woman with a tumor in the anogenital region that grew progressively over the course of 6 months. The giant condyloma acuminatum covered most of the labia majora of the vulva in a continuous pattern. Multifocal condylomas were observed both at the vaginal introitus and at the lateral aspect of the primary tumor.

The patient was tested and had no other sexually transmitted infections. After adequate preoperative preparation, a wide partial vulvectomy was performed. The postoperative course was uneventful.

Adequately planned surgery remains a treatment of choice for patients with giant condylomas.

Keywords: giant condyloma, vulva, low-risk human papillomavirus,

Introduction

Giant condyloma acuminatum, also known as Buschke-Löwenstein tumor (BLT), is a rare sexually transmitted disease of the anogenital region[1,2]. BLT was first described by Abraham Buschke in 1896[3]. Almost 30 years later, Abraham Buschke and his assistant Ludwig Löwenstein described BLT as a penile lesion clinically resembling both common condylomas and SCC, but with a different histological appearance and biological behavior[4-6]. At present, BLT is considered as locally aggressive tumor of benign histological appearance, but with a potential for destructive growth and high recurrence rates. Its development is strongly associated with low-risk human

papillomavirus (HPV) infection[2]. Approximately 90% of genital warts are caused by HPV types 6 or 11[7,8], with HPV type 6 predominating[9]. These HPV types are also found in most BLT cases, demonstrating a key role for these viruses in tumor development[7]. Large condylomas are frequently reported in patients with primary and secondary immune deficiencies[2]. The treatment of choice remains surgery with complete tumor excision and careful follow-up[10,11]. Other treatments options have been described[12], but their effectiveness is uncertain. We report a rare case of vulvar giant condyloma of Buschke-Löwenstein, with a review of the literature.

Case report

We present the case of a 42-year-old female patient who presented to the clinic with complaints of a tumor mass in the genital region. According to her medical history, the tumor has grown progressively over the course of 6 months. Clinically, the tumor resembled a giant condyloma acuminatum, covering most of the labia major bilaterally (Figure 1). Multiple condylomas were observed both at the vaginal introitus and at the periphery of the primary tumor (Figure 2).

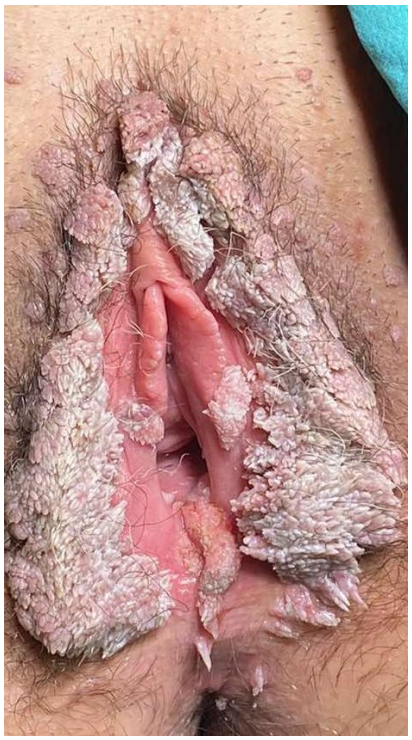

Fig. 1. Macroscopic appearance of bulky cauliflower-like BLT

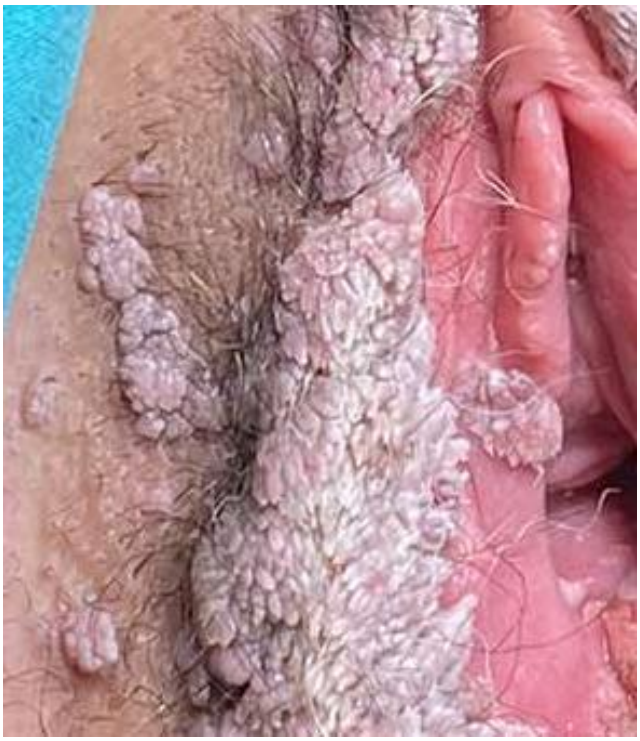

Fig. 2. Macroscopic appearance of BLT with multiple additional acuminate condylomas

Colposcopic examination revealed a normal vaginal mucosa and ectocervix, without any condylomatous lesions. Serologic tests for sexually transmitted diseases were also performed, and the patient was negative for humal immunodeficiency virus (HIV), hepatitis B virus (HBV), hepatitis C virus (HCV), as well as herpes simplex virus (HSV). The patient had normostenic constitution and denied any previous diseases of interest.

A decision was made to treat the patient with a wide partial vulvectomy and to remove the smaller peripheral foci of condylomas using thermal cauterization procedure. The patient was

released from the hospital the next day. Postoperative period was without complications, with complete primary healing of the surgical incisions in 3 weeks.

The histopathologic examination showed epidermal hyperplasia, hyperkeratosis, papillomatosis and koilocytosis (Figures 3-5), with focal mild to moderate dysplasia (Figure 6), consistent with the diagnosis of BLT. The peripheral and deep surgical margins were free of tumor. Additional molecular analysis using polymerase chain reaction (PCR) and probes for low-risk and high- risk HPV types detected a presence of HPV type 6.

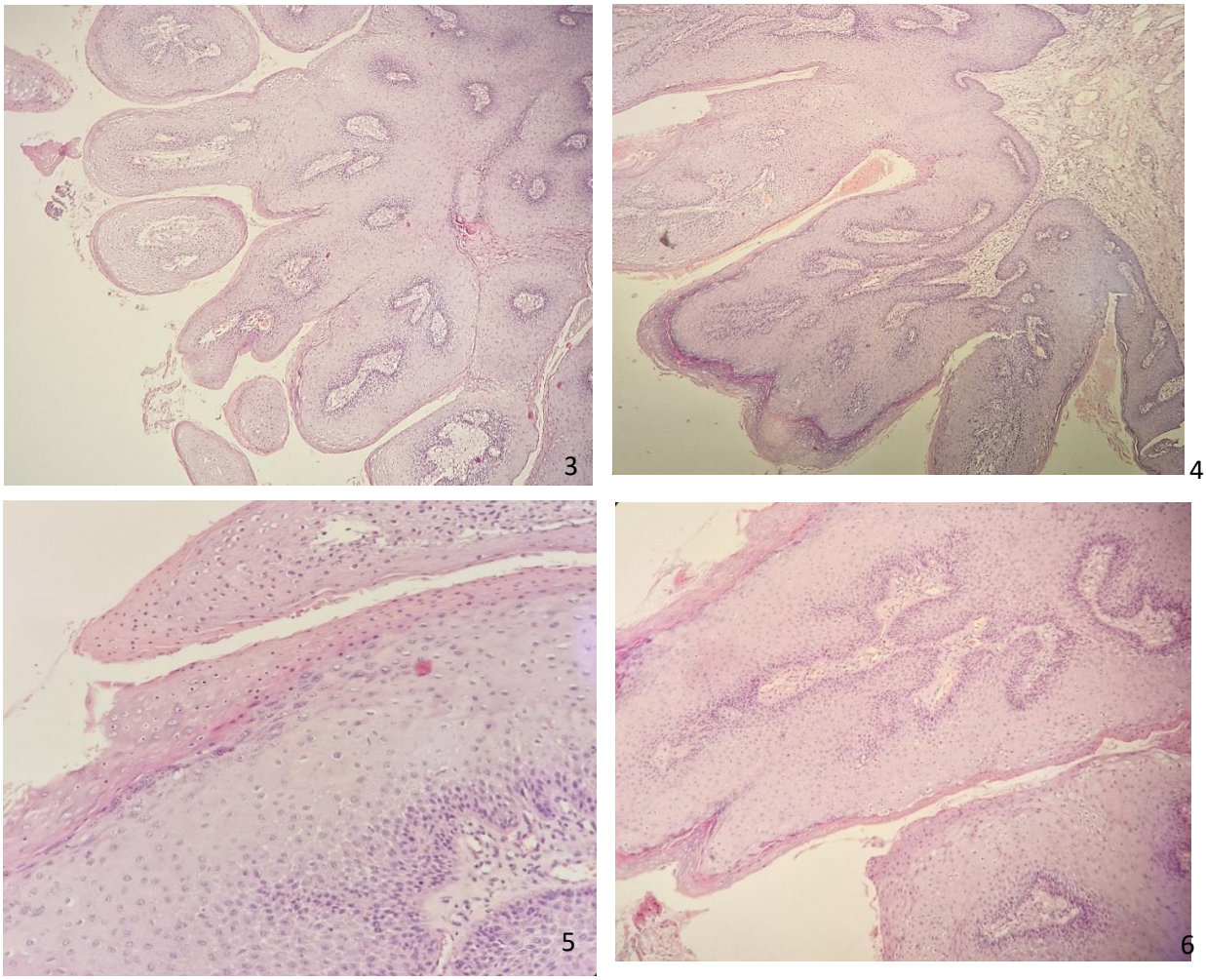

Fig. 3-6. Microscopic appearance of the BLT showing papillomatosis, hyper- and parakeratosis and marked koilocytosis, with epithelial dysplasia

Discussion

BLT is a rare entity sharing histologic similarities to well differentiated verrucous type of squamous cell carcinoma and condyloma acuminatum. However, these tumors are almost always associated with HPV 6 or 11, unlike verrucous carcinomas which are usually HPV-independent[1,13,14]. Its prevalence in the general population is 0.1%[1]. BLT is two to three times more frequent in men, affecting the anogenital region, and most commonly appears between the ages of 40 to 60[2]. These tumors usually occur in immunocompromised individuals[2]. Our female patient was 42 years of age. Acquired immune deficiency due to HIV infection was ruled out with serologic tests.

Malignant transformation can occur in nearly half of the patients, usually without propensity for metastasis[8,15]. The recurrence rate after excision is expected in two thirds of the patients[2,16]. Our patient was lost for follow-up two months after the initial surgery.

The BLT is a slow-growing cauliflower-like mass in the genital or anorectal area, with relatively slow infiltration into deeper tissues[15,17]. The disease starts from a long-standing condyloma acuminatum, which can grow to sizes of more than 10 cm in diameter. In our case, the maximal tumor diameter was 9.5 cm, but several additional smaller condylomas were also present. The tumor usually grows over several years, so unusual clinical presentations of BLT with rapid growth may suggest malignant transformation[18]. Our patient gave anamnestic data of six months duration, and malignant transformation was ruled out histologically.

BLT should be suspected on the basis of patient history and clinical data, but histologic confirmation is mandatory. The BLT histology is characterized by papillomatosis, hyperkeratosis, parakeratosis, acanthosis, and koilocytosis, all of which were observed in our case. Careful histological examination revealed foci of mild to moderate dysplasia or high-grade squamous intraepithelial lesion, without evidence of transformation to SCC.

Optimal recommended treatment for BLT is a wide radical excision, followed by reconstructive surgery[19]. However, for extensive tumors, preoperative chemotherapy or radiotherapy can be used to promote tumor shrinkage, rendering the debulking procedure safer[20].

Conclusion

BLT is a bulky verrucous tumor of the anogenital area with benign clinical behavior. Detailed anamnesis, preoperative investigations, and postoperative histologic examination are crucial for correct diagnosis of these lesions. Carefully planned surgical excision could minimize the risk of recurrence.

Conflict of interest statement. None declared.

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