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Case Report

Presentation of Non-Verrucous Buschke-Lowenstein Tumor. About a Case

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Abstract

We present a rare and unique case of non-verrucous Buschke-Lowenstein tumor, in the form of a very fast-growing vulvoperineal mass, which was completely excised and confirmed by histopathology.

Keywords: *Buschke-Lowenstein (BL); Human Papillomavirus (HPV); Condylomatosis*

Introduction

Buschke-Lowenstein tumor (BL) is an aberrant form of genital condylomatosis, with accelerated growth, verrucous, and very aesthetically disfiguring, caused by the Human Papillomavirus (HPV), generally in immunosuppressed patients [1,2]. Its incidence is very low, but it is associated with a high risk of squamous cell carcinoma between 33 and 66%, in addition to being highly recurrent [3,4].

No other forms of presentation of B-L tumor have been described, other than verrucous, that meet the histopathological criteria to define it, especially due to its rapid tumor development and HPV co-infection.

We present the case of a patient with a rapidly growing vulvoperineal mass with foci of necrosis, which made us think of vulvar carcinoma vs. necrotic condylomatosis of B-L. Its atypical form of presentation, as a semi-hard, non-warty mass, makes us think of other forms of development of this type of genital condylomatosis.

Case Presentation

This is a 42-year-old patient who came to our Service with a painful mass in the vulva that had been developing for 3 months, with rapid growth and color changes for 15 days, accompanied by difficulty urinating and defecating.

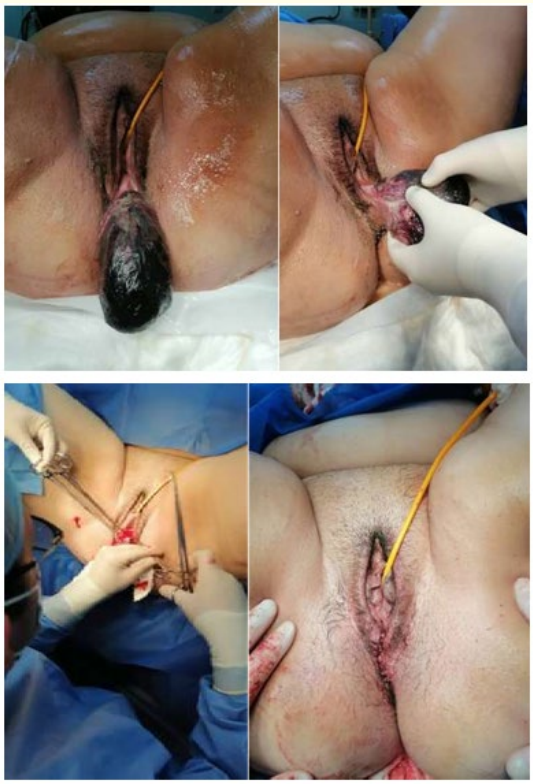
She does not report any significant family or personal history. She has negative cytological reports of cervical intraepithelial lesions in the last 5 years, and does not indicate previous trauma or surgery in the area. She does not report biopsy or treatments for genital condylomatosis. The couple does not report treatments or sexually transmitted diseases or alterations in their external genitalia.

The patient was previously evaluated 6 months ago at another hospital and upon reviewing the medical history of that institution the findings were: presence of a small vulvar mass in the vestibular area, rounded, extending to the vaginal introitus, 2 cm in diameter, soft and not very mobile, hyperkeratotic, and weak aceto. A punch biopsy of the area was performed, which indicated the presence of coilocytes, hyperkeratosis and parakeratosis. There is no report of immunohistochemistry or genotyping of the human papillomavirus. She was diagnosed as probable vulvar lipoma with secondary viral infection.

On clinical examination, a dark mass was seen in the vulva that protrudes from the vaginal introitus in an elongated and bulging shape on the sides, necrotic and shiny, giving it the appearance of an "eggplant", whose measurements are 20 cm long, 10 cm wide and more than 8 cm thick; Smooth surface, semi-soft consistency, mobile and with a base that protrudes from the posterior wall of the vaginal introitus, vestibule and part of the perineum for about 20 mm in length (Photograph 1 and 2). No condylomas or inguinal-femoral adenopathies are observed. Vagina with free course, rough and without lesions. Healthy cervix, aceto-.

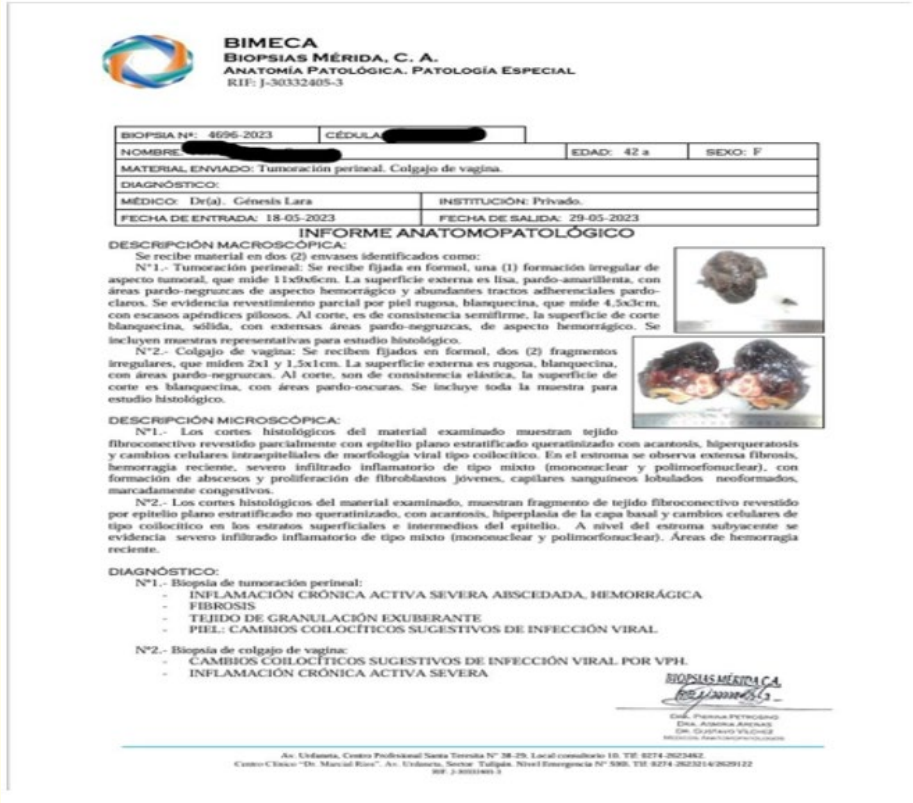
Given the findings, the possibility of an atypical B-L tumor vs. incipient vulvar squamous cell carcinoma was considered as a pre-surgical criterion. An abdominopelvic MRI was requested, but due to financial limitations, it was not performed. It was not decided to perform a bx punch either and due to the surgical accessibility of the mass, it was decided to extract it in block.

The surgical procedure is performed with a low spinal block, then in dorsal lithotomy position, a triangular incision is made from the vaginal introitus to the perineum, progressively removing the entire mass en bloc, with hemostatic control and secondary colpoperineoplasty (Photograph 3 and 4).



Photographs 1-4: *Photographs 1-4 of the tumour mass and surgical result.*

The histopathological report of the piece reports: severe inflammatory infiltrate, abscessed, hemorrhagic and necrotic tissue, fibrosis, exuberant granulomatous tissue, koilocytic changes suggestive of HPV (Photograph 5).



BIMECA
BIOPSIAS MÉRIDA, C. A.
ANATOMÍA PATOLÓGICA. PATOLOGÍA ESPECIAL
RIF: J-30332405-3

BIOPSIA Nº: 4696-2023	CÉDULA: [redacted]		
NOMBRE: [redacted]		EDAD: 42 a	SEXO: F
MATERIAL ENVIADO: Tumoración perineal. Colgajo de vagina.			
DIAGNÓSTICO:			
MÉDICO: Dr(a). Génesis Lara	INSTITUCIÓN: Privado.		
FECHA DE ENTRADA: 18-05-2023	FECHA DE SALIDA: 29-05-2023		

INFORME ANATOMOPATOLÓGICO

DESCRIPCIÓN MACROSCÓPICA:
Se recibe material en dos (2) envases identificados como:
Nº1.- Tumoración perineal: Se recibe fijada en formol, una (1) formación irregular de aspecto tumoral, que mide 11x0x6cm. La superficie externa es lisa, pardo-amarillenta, con áreas pardo-negruzcas de aspecto hemorrágico y abundantes tractos adherenciales pardo-claros. Se evidencia revestimiento parcial por piel rugosa, blanquecina, que mide 4.5x3cm, con escasos apéndices pilosos. Al corte, es de consistencia semifirme, la superficie de corte blanquecina, sólida, con extensas áreas pardo-negruzcas, de aspecto hemorrágico. Se incluyen muestras representativas para estudio histológico.
Nº2.- Colgajo de vagina: Se reciben fijados en formol, dos (2) fragmentos irregulares, que miden 2x1 y 1.5x1cm. La superficie externa es rugosa, blanquecina, con áreas pardo-negruzcas. Al corte, son de consistencia elástica, la superficie de corte es blanquecina, con áreas pardo-oscuras. Se incluye toda la muestra para estudio histológico.

DESCRIPCIÓN MICROSCÓPICA:
Nº1.- Los cortes histológicos del material examinado muestran tejido fibroconectivo revestido parcialmente con epitelio plano estratificado queratinizado con acantosis, hiperqueratosis y cambios celulares intraepiteliales de morfología viral tipo coilocítico. En el estroma se observa extensa fibrosis, hemorragia reciente, severo infiltrado inflamatorio de tipo mixto (mononuclear y polimorfonuclear), con formación de abscesos y proliferación de fibroblastos jóvenes, capilares sanguíneos lobulados neoformados, marcadamente congestivos.
Nº2.- Los cortes histológicos del material examinado, muestran fragmento de tejido fibroconectivo revestido por epitelio plano estratificado no queratinizado, con acantosis, hiperplasia de la capa basal y cambios celulares de tipo coilocítico en los estratos superficiales e intermedios del epitelio. A nivel del estroma subyacente se evidencia severo infiltrado inflamatorio de tipo mixto (mononuclear y polimorfonuclear). Áreas de hemorragia reciente.

DIAGNÓSTICO:
Nº1.- Biopsia de tumoración perineal:
- INFLAMACIÓN CRÓNICA ACTIVA SEVERA ABSCEDADA, HEMORRÁGICA
- FIBROSIS
- TEJIDO DE GRANULACIÓN EXUBERANTE
- PIEL: CAMBIOS COILOCÍTICOS SUGESTIVOS DE INFECCIÓN VIRAL

Nº2.- Biopsia de colgajo de vagina:
- CAMBIOS COILOCÍTICOS SUGESTIVOS DE INFECCIÓN VIRAL POR VPH.
- INFLAMACIÓN CRÓNICA ACTIVA SEVERA

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Photograph 5: *Histopathological report.*

Discussion

The presentation of rapidly growing vulvar masses or tumors is a rarity, considering in the first instance their high risk of malignancy, especially if the patient is premenopausal [3,5,7].

The clinician must evaluate its location, texture, growth and possible causative agents, whether traumatic, infectious, chemical or de novo in the event of a possible malignancy. In this sense, only 3 variants of fast-growing vulvar tumors are described, such as angiomyofibroblastoma [9], which is highly vascularized and when its vessels break, it can generate a hemorrhagic bulging mass, similar to our case; neuroectodermal tumors [10], which are very rare, more common in girls and adolescents, with insidious and highly malignant growth; and finally the B-L tumor, with a showy, warty and deforming growth [3,7]. However, in medicine, not everything is written and the evolution of a pathology varies, depending on the patient's immunological balance and the virulence of the virus.

It is well known that HPV infections cause the formation of condylomas and predispose to vulvar carcinoma in susceptible patients, depending on intrinsic immunogenetic factors and genotypic virulence [3,6]. However, the cytopathic effects of this virus are not associated with accelerated growth of the mucosal-epidermal tissue, and there is no real explanation for why this occurs in cases of B-L; what has been described at an experimental level is the probable activity of the 1-4 receptor of the fibroblastic and epidermal growth factor, due to deregulation of its genes [8].

In our case, the growth of the hyperkeratotic vulvar mass was not in sync with vascular proliferation, which could generate vascular rupture and cell necrosis, giving the tissue that blackish appearance. Hence its symptoms, of progressive pain and mass effect due to its growth, which finally led the patient to seek medical assistance.

On the other hand, our closest diagnosis was a B-L tumor, but we had our doubts because it was not completely verrucous, except at its base, which looked hyperkeratotic, indurated and rough. However, the histopathological result indicated several of its characteristics such as: chronic inflammatory infiltrate, hyperkeratosis, parakeratosis, presence of coilocytes on its surface, fibrovascular papillary formations and low basal mitosis.

In the presentation of this case, we had technical limitations in the management of the presurgical protocol for vulvar tumors for economic reasons, due to the lack of magnetic resonance imaging, immunohistochemistry and viral genotyping, to evaluate within the differential diagnosis the vulvar carcinoma and possible extension. We were guided exclusively by the result of a previous punch biopsy, clinical characteristics of the mass, age and immune condition of the patient.

The treatment of the B-L tumor is generally surgical, but immunotherapy, immunomodulators, antivirals, radiotherapy and chemotherapy have also been described, alone or in combination, according to the presentation characteristics of the lesion.

This case is important due to the presence of an atypical variant of B-L condylomatosis, not described in the medical literature, of clinical utility, for its identification and therapeutic management.

Conclusion

With the presentation of our case, we change the precepts that the B-L Tumor is exclusively verrucous in its presentation, there being variants of the lesion that we must take into account in its diagnosis and clinical and histopathological evaluation.

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