Clinical Case

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Giant Perineal Condyloma Acuminatum (Buschke-Löwenstein Tumour): A Case Report

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Rezumat

Condilom acuminat gigant perineal (Tumora Buschke-Löwenstein): prezentare de caz

Condilomul acuminat gigant, cunoscut și sub numele de tumora Buschke-Löwenstein (BLT), reprezintă o afecțiune cu transmitere sexuală rar întâlnită. În același timp, condilomul acuminat produs de infecția cu HPV (Human Papilloma Virus) devine una dintre cele mai comune infecții transmise pe cale sexuală ce afectează regiunea perineală. În aceste situații, opțiunea terapeutică de prima intenție este excizia chirurgicală. Obiectivul acestui articol este de a prezenta un caz în care tumora a atins dimensiuni gigante, ce afectează calitatea vieții și de a accentua importanța unei intervenții chirurgicale largi, având în vedere rata ridicată de recurență și potențialul semnificativ de transformare malignă a BLT.

Cuvinte cheie: gigant, condilom, Buschke-Löwenstein, vulvoanal, HPV, neglijat

Abstract

The giant condyloma acuminatum, also known as Buschke-Löwenstein tumor (BLT), is a rare sexually-transmitted disease. Moreover, the condyloma acuminatum produced by Human Papilloma Virus (HPV) infection becomes one of the most common sexually-transmitted infection which affects the perineal region. Under such situations, the first therapeutic option is surgical excision. The objective of this article is to present a case in which the tumor has reached giant dimensions and also to stress the importance of an in toto resection, taking into account the high rate of recurrence and the significant potential of malignant transformation of BLT.

Key words: giant, condyloma, Buschke-Löwenstein, vulvoanal, neglijat

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Introduction

The giant condyloma acuminatum, also known as Buschke-Löwenstein tumour (BLT), is a rare sexually-transmitted disease (STD) (1), with an incidence of approximately 0.1% in the general population (2), whose main risk factor is the infection with the Human Papilloma Virus (HPV) (3).

BLT is characterized by an invasive growth, a high rate of recurrence to the treatment and likely potential of malignant transformation (squamous cell adenocarcinoma) (1). This is a

cauliflower-like tumour of significant dimensions, with a slow development in time (3). From a histopathological point of view, it presents benign characteristics, although from a clinical perspective, it might determine signs of malignancy (3), secondary to the pressure put on nearby tissues, to the lack of spontaneous regression and to the high rate of recurrence despite a correctly administered treatment with medication.

Case report

A 52-year old woman presents with a giant tumour lesion in the vulvo-perineal region, skin pallor and tendency to fainting.

The patient is now for the first time in a medical specialty facility although the lesion appeared approximately 10 years before and it gradually grew in dimensions (more accelerated in the last year).

The clinical examination revealed an ulcerative-vegetative tumour, fetid, hemorrhagic in the vulvo-anal region, of roughly 25/15 cm, which extended to the root of the left thigh and which covered the anal orifice, while the digital rectal examination was impossible to perform (Fig. 1). The local superficial lymph nodes could not be found at palpation.

The lesion was significant for a Buschke-Löwenstein Tumour (Fig. 1).

The pathological history of the patient did not reveal chronic diseases or the administration of some medications which might have suppressed the function of the immune system.

The laboratory tests reveal severe microcytic, hypochromic anemia with a haemoglobin value of 6.14 g/dl, while the rest of the investigation was unchanged, including the inflammatory tests.

The ELISA test for detecting the HIV1/HIV2 antibodies was negative. The X-ray on the pelvis did not reveal osseous invasion in the vicinity, and the ultrasound did not reveal any secondary determination.

The patient receives 7 isogroup isoRh blood transfusions, eventually the haemoglobin reaching a value of 10.16g/dl, close to its normal value (12 g/dl). Given the urgent character of the surgical intervention (an over infected and hemorrhaging giant tumor led to severe anemia, hard to correct because of the group and Rh particularities of the patient), we preferred the perprimam excision of the whole tumor until healthy tissue was reached (instead of an excisional biopsy), and we shall adapt the therapeutic management according to the pathological result.

After receiving the written consent of the patient, surgery is performed and the giant tumour is totally excised by means of an electric scalpel, until healthy tissue is reached in a gynaecologic position under general anaesthesia. During the surgical intervention, 6 other tumours similar from a macroscopic point of view were excised. They were smaller in dimension, with a bilateral labial and perianal location.

After the preparation of the skin edges, the per-primam suture with rare threads to the integument was performed (infected tumour). At the same time, a wide spectrum antibiotic was administered intravenously (both pre- and postoperatively), along with antialgic and anti-inflammatory treatment, and the

Figure 1. Lesion view on diagnosis

postoperative evolution was slowly favourable. The wound is cleaned and dressed on a daily basis.

Despite its impressive dimensions, the histopathological outcome was papillary condyloma acuminatum, with no signs of invasion, dysplasia or any malignant transformation (Fig. 2).

Wound dehiscence occurred in approximately 15 days after surgery, probably due to the strain of the integument during defecation and due to left thigh movement, but also due to the fact that the tumour was infected.

The patient is washed and dressed on a daily basis until the reach of a granular eutrophic wound ready for grafting (Fig. 3).

Then, the patient undergoes autologous skin grafting with a graft harvested from the left thigh (Fig. 4).

The postoperative evolution is favourable, with the vascular and metabolic integration of the graft, with no further

Figure 2. Microscopy of the vulvo-perineal Buschke-Löwenstein Tumor

Figure 3. Granular eutrophic wound

complications. The patient is discharged after she is surgically cured. The check-ups were done on a weekly basis in the first month and then on a monthly basis and they revealed local recurrence only after 3 months from the discharge date.

Discussion

This case ranks among the few condilomas with giant development which evolve like tumours occurred in the vulvo-perineal region and which lead to the significant degradation of the patient's life quality due to their impressive dimensions. Also, repeated haemorrhages led to the occurrence of severe chronic anaemia, more and more difficult to tolerate by the patient. Vicinity to the external anal orifice led to tumour infection along with the occurrence of a heavy smell.

According to the data in the literature, the condyloma acuminatum can be treated through various non-surgical methods: the application of a trichloroacetic acid solution 80-90% or podophyllotoxin solution 0.5%, radiotherapy, chemotherapy, immunotherapy and cryotherapy with liquid nitrogen, but their efficiency is controversial (6,7,8,10). Topical application of podophyllin in case of Busche-Lowenstein tumors is not recommended due to its bad results, even if it has good results on ordinary acuminatum condyloma (19).

There were cases in which the 5-fluorouracil therapy was attempted, but the outcomes were insignificant, later on being used together with cryotherapy (9).

Topical immunomodulators (Imuquimod cream 5%) or antivirals with topical application or intralesional injection

Figure 4. Reconstruction of the large wound defect with skin graft

(Cidofovir) can also be used. Other means of nonsurgical treatment, with low utility and local application are: salicylic acid soil 4-5%, glutaraldehyde 10-20%, bleomycin and interferon alpha intralesional injections.

Surgical management consists in classical surgery, electro-surgery (electrocoagulation, radiofrequency and carbon laser surgery) or both.

Electro-coagulation with an electric scalpel is still in use for small dimension condilomas, but this procedure has a significantly high risk of relapse.

Another means of treating giant condylomas acuminatum is the use of carbon laser, used after the initial treatment with imiquimod or after surgical excision (18).

Although there are many therapeutic options, they are not more efficient than the rest, efficacy which couldn't be supported. Because there are no gold standards in managing BLT cases, surgery remains the most common method of treatment. The data in the literature show satisfactory results through the use of either monotherapy or combined therapy (17).

In this case, these methods were not taken into account due to the impressive dimensions of the tumour, and the only option was the surgical excision.

At present, local radical excision is very important (4, 5, 13, 14) due to the risk of malignant degeneration which completes the reconstruction either at the same operative time or subsequently (plasty with detached skin or, in case of bigger defects, both integumentary and soft underlying parts – the use of flaps with own blood supply (11,19). Adjuvant therapy such as radiotherapy or immunotherapy might have favorable effects, but their effectiveness is still under study (19).

In this case, due to the impressive dimensions of the tumor, the good immune state of the patients and also the costs and availability, the only valid option was the surgical excision along with the defect coverage, and in a subsequent meeting

with the detached skin graft. The complications that may appear after the surgical treatment include: local recurrence, hemorrhage, wound dehiscence, infections, pain, functional disabilities and malignant degeneration.

The infection of the wound due to proximity to the anal orifice might suggest, as a temporary solution, the creation of an artificial anus at the level of the sigmoid colon (19).

Local periodical and long-term monitoring is indicated due to the high incidence of recurrences in case of Buschke-Löwenstein tumours.

In this case, the surgical treatment was postponed for a while due to the difficulty in administering compatible blood to the patient, since her blood type is rare (0, negative Rh).

Benign anal-genital lesions (condyloma acuminatum) are associated with nononcogenic strains of the HPV (with a low risk): 6 HPV and 11 HPV genotypes, seen in more than 90% of all cases (15). In our patient's case, the test to detect the infection with HPV was not run, since it was meant to be performed subsequently. In most cases, the female patients with Buschke-Löwenstein tumours have an immunodeficiency state, but in this case, the ELISA test for detecting HIV1/ HIV2 antibodies was negative, and the patient did not have any other pathologies or conditions that might have indicated immunodeficiency. Therefore, the characteristics of this case are: the impressive dimension (occurred in the context of prolonged negligence of the patient), the occurrence of Buschke-Löwenstein tumour in a patient without an obvious immuno-deficiency, its occurrence in a female patient, since the data in the literature indicate a predisposition for male patients (16).

Conclusions

Negligence, shame, fear induced by a possible malignant lesion often lead to a delayed arrival to the doctor, which might affect the life quality, might increase the risk of malignant degeneration and the likelihood of tumour growth to monstrous dimensions. These might lead to hemorrhagic compressive and infectious complications, to a more difficult surgical treatment, with an increased risk of relapse and with possible complications and postoperative sequelae.

The in toto surgical excision, associated with the autografting of the resulting defect and close postoperative monitoring, represents the ideal solution for the treatment of giant Buschke-Löwenstein tumours.

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