

A Rare Case of Perineal Giant Condyloma Acuminatum

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Abstract: Giant condyloma acuminatum, also known as Buschke-Löwenstein tumor (BLT) is a very rare sexually transmitted disease that affects the ano-genital region. This tumor has a locally destructive behavior, a high recurrence rate and occasional transformation to squamous cell carcinoma. Risk factors include anoreceptive intercourse, HIV and immunosuppression. We present a case of perianal BLT and discuss about the nature and treatment of this disease. Our aim is to emphasize some basic points in the management of giant perianal condyloma acuminatum. The patient with BLT must be very carefully assessed clinically and imagistic investigated in order to detect the tumor visceral invasion and to establish the extension of the surgical procedure. Wide perineal excision with negative margins is the best surgical choice if the anal canal is not involved. The radical pelvic surgery is indicated only in patients with proved visceral invasion. Excision is mandatory even in very small condylomas to prevent BLT later.

Keywords: giant condyloma acuminatum, Buschke-Löwenstein tumor, wide perineal excision

INTRODUCTION

Giant condyloma acuminatum (GCA) is a slow-growing, large, cauliflower-like tumor with a locally destructive behavior that typically appears in the anogenital region. It was originally described as a penile lesion by Buschke in 1896 and Lowenstein in 1925 [1]. The first description of anorectal GCA was by Dawson *et al.*; [2] in 1965. GCA is a rare lesion tending to present in the fifth decade with a 2.7: 1 male: female ratio. For patients <50years, this ratio is increased to 3.5: 1 [3]. Risk factors include HPV [6, 11] anoreceptive intercourse, HIV and immunosuppression.

Common clinical signs that can be found in such patients are bleeding, lymph node enlargement, usually inflammatory, abscess and perianal fistula [4, 5]. The loco-regional extension must be carefully assessed to establish the therapeutic strategy. In respect to this, abdominal and pelvic computed tomography and magnetic resonance imaging, and intra anal ultrasound-endoscopy are useful [6].

Histologically it is characterized by aggressive down growth into underlying dermal structures [7, 8]. A complex histological pattern may exist with areas of benign condyloma intermixed with foci of atypical epithelial cells or well differentiated squamous cell carcinoma.

Besides pathogenesis, there are also controversies on the BLT treatment [9]. The majority of authors agree that surgery is the treatment of choice and is efficient in the early stages of the disease [10].

Excision must be wide [11, 12]. Lymph node dissection is indicated only in cases of suspected malignant transformation [4]. Some authors prefer abdomino-perineal amputation, but others support the conservative surgery as the best choice, mainly in terms of the patient's quality of life [13]. The high local recurrence rate after excision, of about 50-60%, has suggested the need of pre- and postoperative use of radiation and chemotherapy [14, 15]. There are reports that loco regional control of recurrent forms was achieved with combined chemo radiotherapy [16, 17, 18]. There are also rare reports of primary radiotherapy producing complete tumor regression [21]. Post-treatment monitoring is strongly recommended [4]. We present a case with perianal BLT, and discuss some controversies about the nature and treatment of this disease. Our aim is to emphasize some basic points in the management of giant perianal condyloma acuminatum.

CASE REPORT

45 year old male patient, newly diagnosed HIV (+), without anti-retroviral therapy, viral load of 57,000 RNA copies/ml, CD4 T lymphocytes: 418. Consultation for a mass at the base of the penis and pubic region of 6 months duration. Onset was insidious and progress was gradual. Physical examination revealed a cauliflower-like tumor, involving the base of penis, pubic area and left side scrotum.(figure 1).Giant condylomata acuminata is diagnosed . The decision was taken to perform surgical excision of the lesion with negative margins. (Figure 2).complete excision was achieved. Split skin graft is performed to cover the defect, the donor area being medial side of ipsilateral thigh. Patient

recovery was uneventful. Histological analysis shows hyperplasia and acanthosis of the perianal skin, with inflammatory reaction of the adjacent rectal mucosa, without atypias, compatible with giant condylomata acuminata. Six months following the surgery there was no recurrence.



Fig-1



Fig-2

DISCUSSION

Buscke-Lowenstein tumor is a rare entity, with an incidence of 0.1% in the general population [17, 18, 19]. Predominantly affects men, with few reports in women, being more common during pregnancy [17, 18, 20, 21, 22]. Presents rates of up to 56% of malignant transformation to squamous cell carcinoma, 66% recurrence and 20% mortality, with fatal cases only in recurrences [17-19, 23, 24]. This disease has been considered an intermediate step between squamous carcinoma and condyloma acuminata or a benign entity in itself with malignant behavior [18, 19, 23-25]. There are little known clinical characteristics and imaging, and there is no agreement on handling/management. Risk factors described are HPV subtype infection [6, 11, 16, 18], immunosuppression (HIV infection, use of corticosteroids, immunomodulators, diabetes mellitus) sexual promiscuity and co-existence of condylomas [18, 19, 22]. The most frequent locations in males are the

penis (81-94%) and in females the vulva (90%), secondly in both sexes is the perineum [19, 24, 26]. Lymphadenopathy associated with this lesion are mostly reactive to the lesion or super infection, they rarely correspond to metastasis [19, 24, 26, 27].

Clinically these lesions often present with similar findings, and can be distinguished by histological examination [30–31]. At histology, GCA differentiates from other lesions by its thick stratum corneum, marked papillary proliferation, and tendency to deep invasion, with displacement of the surrounding tissues [29]. These same features, seen also in verrucous carcinomas, led several authors not to recognize a distinction between verrucous carcinoma and Buschke-Löwenstein tumor [28]. However, GCA does not present histological evidence of malignancy, such as infiltration of basement membrane, lymphatic invasion, angioinvasion or distant metastases [32]. Despite this, GCA can coexist with verrucous carcinoma or squamous cell carcinoma in up to 50% of patients [29, 33].

So far, no definitive therapeutic strategy has been established [5]. The management of BLT must be implemented knowing the pelvic tumor extension which needs clinical and imaging exams.

The majority of authors agree that surgery is the treatment of choice and is effective especially in the early stages of the disease [15]. Wide local excision remains the mainstay of therapy, that can be followed, if it is necessary, by delayed split thickness skin grafts [1, 11, 25, 9]. There are some controversies between radical abdomino-perineal amputation and conservative surgery supporters [13].

We think that wide perineal excision with histopathological margins control is the best surgical choice in the treatment of BLT if the anal canal is not involved. The radical pelvic surgery is reserved only for patients with provable visceral invasion. Wide resection is indicated even in rare cases of benign nature BLT because of the high risk of carcinomatous transformation and the great discomfort produced by the giant perineal tumor. We also emphasize that surgical excision is mandatory in all cases of BLT when it is technically possible and also in all condylomas, even in very small ones, in order to prevent their BLT transformation.

There is no consensus about the use of pre- and postoperative oncologic treatment in BLT. Some authors recommend that reduction of the tumoral mass through radiotherapy or chemotherapy to precede surgical excision [4, 15]. Others assert that radiotherapy is rarely used, usually after an incomplete excision, in recurrences and when excision is not recommended.

There are rare reports of primary radiotherapy producing complete tumor regression [21]. Radiotherapy has been suspected of being responsible for the alteration of BLT into anaplastic carcinoma [15, 16, 26].

Some authors reports the use of imiquimod and others present a case of BLT successfully treated using carbon dioxide laser vaporization and systemic interferon therapy after failure of 3 surgical excisions [2, 27-30]. The patient must be long time closely followed-up [4,31]. We think prospective studies are necessary to further define the nature and treatment of this very rare disease [32].

CONCLUSION

The optimal treatment for Buschke-Löwenstein tumor is still being debated because of the lack of a consistent series of patients. Despite this, at the moment, wide radical excision with plastic reconstruction of skin defects seems to be the best choice. Adjuvant therapies, such as radiotherapy and immunotherapy, may achieve good results, but their effectiveness is still uncertain.

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