

Buschke Löwenstein Tumor of the Right Lower Abdominal Wall: Case Report and Review of the Literature

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Abstract: Buschke-Löwenstein tumor (BLT), known as giant condyloma acuminatum (GCA), is a very rare disease that typically appears as a penile lesion but can although appear in the anogenital region, bladder, vulva, scrotum and sacrococcygeal area as well.

Despite of its histologically benign signs, a high recurrence rate, invasiveness and destructive growth characterizes this rare disease as clinically malignant.

Malignant transformation into verrucous carcinoma (VC) and squamous-cell carcinoma (SCC) have been described as well.

Many treatment modalities inclusive neoadjuvant radio-and chemotherapy and topical treatment have been reported but due to lack of controlled studies no treatment can be recommended.

We present a case of Buschke-Löwenstein tumor involving the right lower abdominal wall of the colostomy region at a 71 years old male.

To our knowledge, we first describe a case report of GCA involving abdominal wall at the colostomy region, successfully treated by wide radical excision and plastic reconstruction.

Keywords: Buschke-Löwenstein tumor, Giant condyloma acuminatum, surgical resection, neoadjuvant therapy, squamous cell carcinoma.

INTRODUCTION

The giant condyloma acuminatum (GCA) also known as Buschke Löwenstein Tumor (BLT) was first described as a penile lesion in 1925 [1]. It is a cauliflower like tumor involving external genitalia and perianal region. The anorectal GCA was first described by Dawson *et al.* [2].

The GCA is pathologically a rare entity. Some authors classify the GCA as a low grade squamous cell carcinoma (SCC) with a minimal risk of metastasis: a verrucous carcinoma (VC) [25] and others subscribe GCA between normal viral warts and VC [5].

Malignant transformation has been reported in 30-56% of all cases and the male-female ratio is 3,5 :1 [3]. The human papilloma virus (HPV) is implicated in the pathogenesis of GCA. Risk factors include immunosuppression, anoreceptive intercourse and HIV.

Many sporadic reports have been published to describe various surgical and non-invasive treatment options. Either radical surgery, topical application,

radio-chemotherapy or immunotherapy has been used [3-5].

CASE REPORT

We present the case of a 71 years old heterosexual male with a history of rectal cancer (UICC Stage III) after total mesorectal excision in 2004 and after following combined adjuvant radio-chemotherapy. In June 2011, he had a re-laparotomy to clarify the histology of a retroperitoneal metastatic area and the same at the pelvic wall. Histopathologically cancer recurrence has been excluded. Hartmann's Procedure with a colostomy was performed at the re-operation.

In October 2012, this patient was admitted to our clinic presenting a cauliflower like tumor mass at the colostomy in the right lower abdominal wall. In the current history he noticed a small warty lesion with occasionally bleeding 10 months ago. In the last 2 months, the care of the colostomy worsened and the bleeding episodes became shorter. No signs of obstruction were detectable at the time of admission. No lymph nodes were palpated in the inguinal area bilaterally.

No evidence of an immunological deficit was detected at the admission. The serology of Hepatitis B and C was negative.

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Except a mild anemia, the laboratory results showed no abnormalities. A chest x-ray showed no signs of suspicious lesions and the preoperative colonoscopy was without unobtrusively as well.

Afterwards, surgical procedure was performed by excising a wide local skin and subcutaneous area of 1,5 -2 cm radial and a colonic resection of about 8 cm as well as a new plastic reconstruction using a skin flap. Then, a colostomy was placed at the same position.

The histological examination of the tumor showed bands of proliferating undulating squamous epithelium.

It was marked with hyperkeratosis, acantosis and a sharply defined margined lymphocytic infiltrated hypergranulosis. Koilocytes were found in the superficial areas. No cell dysplasia was visible (Figure 2).

The postoperative course was proceeded without a complication and primary wound healing.

Neither topical therapy, nor a radiochemotherapy or immunotherapies were applied postoperatively.

Finally, examinations of the wound after 2 weeks, after one, three, six and twelve months showed vital



Figure 1: Macroscopic view (after tissue fixation) shows cauliflower like tumor (BLT) boarding on the colonic mucosa of the colostomy.

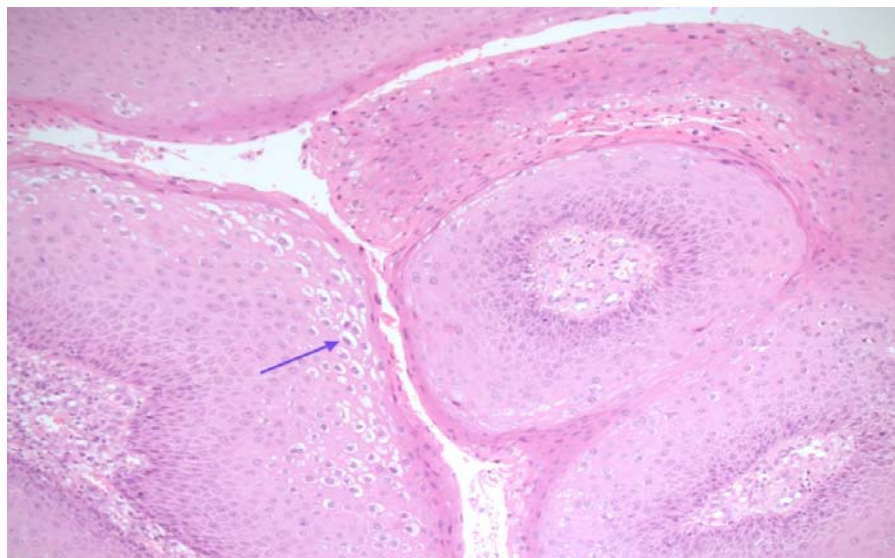


Figure 2: Hematoxylin and eosin stain with typical koilocytes (arrow).

skin and colostomy and no evidence of tumor recurrence was seen.

DISCUSSION

The case presented emphasizes the successful surgical resection of a giant condyloma accuminatum, at a colostomy site with infiltration of the abdominal wall.

As far as known, it is the first description of the uncommon localisation concerning this rare illness.

Mostly, the GCA is found in the perianal region. Some other locations such as the sacrococcygeal pilonidal sinus tract, the cervix, a rectal and scrotal growth or bladder involvement have been described in the literature [5-8, 14].

The etiology of GCA still remains uncertain. The HPV known to cause condyloma acuminatum, also suspected to induce these tumors [9]. Pathogenetically, a low-risk HPV types 6, 11 and high-risk HPV 16 and 18 have been found. Their functions in the potential malignant development have not been clarified [10]. Either the presence of HPV is a coincidence nor a cofactor in carcinogenesis [4, 26].

Creasman *et al.* reported about 6 cases from 30 GCA that developed a squamous cell carcinoma. The authors support the hypothesis that the GCA represents an intermediate lesion in pathology of squamous cell carcinoma [4].

The thick stratum corneum, marked papillary proliferation, and tendency to deep invasion with displacement of surrounding tissue are the main histological signs of GCA [3]. Similar features are also seen in verrucous carcinoma and some authors do not differentiate between a Buschke Löwenstein tumor and a verrucous carcinoma. However, GCA does not show signs of malignancy [12]. These were detected in our histopathological examination. GCA can coexist with verrucous or squamous cell carcinoma in up to 50% of the cases [3, 13].

GCA is often associated with patients suffering from immune defects like HIV-infected patients or patients undergoing immunosuppressive treatment [5, 14]. Piepkorn *et al.* [11] described the development of GCA during cyclosporine therapy. The etiology in our patient has not been clarified. We consider the negative influence on the immune system after adjuvant radio-chemotherapy, despite no immune deficiency was detected.

Many different therapeutical approaches were included in the treatment of GCA. Because controlled studies are missing, no standard therapy can be recommended. Individual management based on surgical experience and single case reports should be considered in the targeted treatment of this rare disease [15].

At this point, wide local excision remains the treatment of choice [16].

Many other none-invasive options have been reported. Topical treatment with podophyllin successfully described in the management of ordinary condyloma acuminatum. But in the treatment of GCA, podophyllin was observed with high recurrence rates [17].

Immunotherapy has been successfully reported by several authors in small numbers of patients [18, 19]. An autogenous vaccine prepared from patient's tissue is not a treatment option at its own. It can be useful in the reduction of a tumor bulk prior to surgery.

Chemotherapy alone plays an uncertain role in therapy of these lesions. Certainly, a combination of chemo-and radiotherapy may downsize these lesions to become suitable for surgical resection. Neoadjuvant therapy based on 5-fluorouracil/mitomycin and 5-fluorouracil/cisplatin has been shown as being successfully „downstaging“ in few cases [20,21]. Combined radio-and chemotherapy is the most preferable therapy if surgery is impossible [22].

Despite of this, the results of radiation therapy still remains controversial. Reportably, the behaviour of the tumor turns to be more aggressive after the radiation and there were up to 10% of anaplastic transformation after therapy [23,24].

All of none-invasive modalities mentioned describe reduction of the tumor mass preoperatively, but it is not very effective in reducing the persistent high recurrence rate up to 60% [3].

CONCLUSION

We agree that the early surgical approach with wide resection should be the first choice in the management of GCA as it provides the best chance of survival. Because of the lack of controlled studies, no general recommendation in the treatment of GCA can be made. Neoadjuvant therapy or initial topical therapy should be considered to reduce the tumor mass and improve local situation prior to surgery.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

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