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REVIEW ARTICLE

PERINEAL GIANT CONDYLOMA ACUMINATUM OR BUSCHKE-LOWENSTEIN TUMOR: A REPORT OF TWO CASES

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ABSTRACT

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Key words:

Buschke-Lowenstein Tumor, Giant Condyloma Acuminatum, Humanpapillomavirus, Perinium, Sexually ransmitted disease. A study was undertaken Buschke-Lowenstein Tumor (BLT), also known as Giant Condyloma Acuminatum, is an uncommon and locally invasive tumor primarily affecting the anogenital region, often associated with human papillomavirus (HPV). This paper reports two cases of BLT successfully managed with staged operative procedures. Surgical excision, followed by secondary healing, yielded favorable outcomes without complications. BLT management remains a clinical challenge due to its size and recurrence rates, highlighting the importance of a multi-disciplinary approach and further research to enhance treatment strategies. These cases contribute to the understanding of BLT and emphasize the need for improved interventions to enhance the quality of life for affected individuals.

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INTRODUCTION

Buschke-Lowenstein tumour (BLT), also known as Giant Condyloma Acuminatum, is a very rare, locally invasive tumor. It is a sexually transmitted disease that affects the anogenital region.(1-3)It was first described by Abraham Buschke and Ludwig Loewenstein in 1925. Although it is a well-differentiated, benign tumor, its management is often challenging due to the size, probable local invasion, and recurrence rates which may result in poor quality of life with physical and psychological morbidity. Progression to a giant condyloma makes sexual intercourse impossible and mitigates against proper hygiene of the perineum. Although there is no well-defined treatment protocol for BLT, many medical and surgical treatment options have been described in literature. We report two cases of patients with giant condyloma successfully treated by staged operative procedure.

CASE REPORTS

Case 1: A24-year-old female, was referred from the department of dermatology with a one-year history of a growing mass in the vulval and perianal area with pain and discomfort. She had not received any treatment previously. The mass had progressively increased in size and was making intimate personal hygiene increasingly difficult to maintain. She denied sexual promiscuity. She was diagnosed with retroviral disease since birth and was receiving ART.

On examination, there was a cauliflower-like growth on her both labia majora obliterating the pudendum with few satellite lesions on perianal region and seedling lesions on labia minora while clitoris and periurethral region were spared. The lesion was fleshy, sessile, and slightly friable in areas with some bleeding and foul-smelling, purulent exudate on the surface. Grossly, the lesion measured 10×5 cm with a central thickness of 4 cm (Figure 1 and 2). Per-speculum examination revealed normal vagina and cervix and inguinal lymph nodes were not palpable. Routine laboratory investigations were within normal limits except seropositive status for retrovirus. CD4 counts were 200 cells/µL. The biopsy of lesion showed moderate degree of dysplasia of the epithelium with koilocytosis, acanthosis and parakeratosis. After obtaining written consent, partial surgical excision was done under spinal anaesthesia with an electrocautery removing the entire mass. The wound was left open for secondary healing.Twice daily sitz bath with antiseptic solution combined with analgesics (Paracetamol) and antibiotics (Amoxicillin with clavulanic acid) were advised. This led to healing in two weeks. Histopathology confirmed features consistent of Buschke-Lowenstein tumour. During the second follow-up visit after three months, residual lesions were observed on the labia majora, labia minora, clitoral area, and perianal region. These residual lesions had grown larger compared to her previous visits. After 3 months second stage procedure was planned for her with removal of residual lesions with electrocautery (Figure 3A and 3B). The entire wound was left open to heal by secondary intention. These conservative therapies went without complications.



Figure 1: Extensive Cauliflower like growth on labia majora with few satellite lesions on labia minora and perianal region with obliteration of pudendum- (Case-1)



Figure 2. Few satellite lesions on perianal region and seedling lesions on labia minora with sparing of clitoris and peri-urethral region (Case-1)



^{3A} ^{3B} Figure 3. AResidual lesions noted over labia majora, labia minora,clitoral area& perianal region which were increased in size after 3 month follow up (Case-1)

She is now in the 1st year of follow-up (every 3 months) and presents no residual lesions or recurrence. Furthermore, the functional results were excellent with no evidence of anal canal incontinence, and the cosmetic results were satisfactory.



Figure 4 - Follow up image after 1 year of second surgery (Case-1). No local recurrence and patient was satisfied with the appearance



Figure 5. Pre-operative image showing lesion measured 6 X 4 cm with central thickness 2 cm on the right labia majora (Case-2)



Figure 6. Follow up image showing recurrence after 1 year (Case-2)

Case 2

A 47-year-old female presented with five-year history of a growing mass on the anterior half of the right labia majora and perianal region with excessive foul smelling discharge and had associated pruritis . Patient had no history of any treatment in the past for the same condition andno history of retroviral disease. Grossly, the lesion measured 6 X 4 cm with a central thickness of 2 cm on the right labia majora.Surgical excision was done by electrocautery and the wound was left open for secondary healing. Patient did not follow up as advised after discharge. Patient came after 1 year of surgery with recurrence but declined further treatmentbecause she did not experience any functional problems such as itching or foul-smelling discharge.

DISCUSSION

Approximately 70 HPV genotypes can infect the skin and mucous membranes, with a preference for transitional mucous membranes where they can cause pseudo-epitheliomatous proliferation, tissue dysplasia, and neoplastic transformation.(5) Genotypes 6, 11, 42, 43, and 44, found in more than 90% of cases, produce condylomatous lesions and are associated with a low oncogenic potential. On the contrary, genotypes 16 and 18 are associated with a high risk of dysplasia and squamous cell carcinoma (6,7). The Extensive form of BLTs is much less frequent and is reported as clinical cases or short series in the literature. Evolution to the extensive form is favored by chronic local irritation, poor personal hygiene, and immunosuppression (HIV, immunosuppressive treatment, diabetes, chemotherapy) Association with other sexually transmitted infections is common (HIV, syphilis, and chlamydia). (2,6,8).

BLT affects all ages, with the majority of cases occurring between the 4th and 6th decades of life.(2) It is a tumor with both exophytic and endophytic growth that can deeply infiltrate structures, leading to the suspicion of malignant transformation to microinvasive carcinoma or in other cases to invasive keratinizing squamous cell carcinoma(4,6). Balla et al reported the cases of two male patients aged 45 and 57 years, who presented with perianal and buttock BLT for which large excisions were performed with satisfactory outcomes. (8). Zhang et al presented the largest case series of 38 cases of BLT and characterize its unique clinicopathologic features. The study indicated that certain histologic features such as dyskeratosis, neutrophilic micro-abscess, and abnormal mitosis. They concluded that complete surgical excision is considered the preferred initial therapy with good prognosis. (9). Spînu D et al discussed various aspects of this rare and aggressive form of genital warts, and concluded that surgical excision remains the "gold- standard" treatment. A detailed histological exam of the lesion is mandatory, because microscopic excision (with tumor-free margins) presents a decreased recurrence rate compared to macroscopic resection. Minimally invasive surgery can be successful in small-sized lesions and recurrence cases. (10)

Surgical treatment is the gold standard, although it can be disfiguring. The surgical excision must be large and must involve all macroscopic lesions with adequate resection margins. In our two patients, the lesions were non-invasive and easily excised with a electrocautery followed by hemostasis. In the absence of invasion and obvious degeneration, surgical excision is sufficient in most cases and healing by secondary intention is the most widely used method in the literature. It involves twice-daily care (by sitz bath with antiseptic) with a healing time of more than 2 months and sometimes predisposes to cicatrization.(7,8,9) Flap coverage shortens the healing time and improves the aesthetic results.(4,18). Recurrence may occur in 50% of cases, due to incomplete excision or the progressive development of microscopic lesions.(9,11) The average recurrence time is 10 months. Topical adjuvant therapy is proposed in cases of remaining lesions with variable results. However, recurrence occurs in 67% of cases after medical treatment(11). The presented cases highlight treatment outcomes using operative procedures. Surgical excision was the primary approach, removing the entire mass in both cases. Healing by secondary intention, aided by sitz baths and antibiotics, led to favourable outcomes. The cases demonstrated good functional and satisfactory cosmetic results without complications.

CONCLUSION

This study presented two cases of Buschke-Lowenstein Tumor (BLT) or Giant Condyloma Acuminatum successfully treated with staged operative procedures. BLT, a rare and locally invasive tumor linked to HPV, poses complex challenges due to its size and recurrence rates.

The good early results followed by recurrence in the second case demonstrates the virulent nature of the disease and the need for counselling of the patients about the necessity of repetitive surgery. Experiences of other surgeons and patients will help increase our knowledge of the progression of the disease. Surgical excisionfollowed by secondary healing led to favourable outcomes. BLT management remains a clinical challenge, emphasizing the need for a multi-disciplinary approach and further research to enhance treatment strategies and improve the quality of life for affected individuals.

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