The Conundrum of an Accumulating Acuminatum

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Conflict of interest: None declared

Patient: Male, 70
Final Diagnosis: Chronic condyloma acuminatum
Symptoms: Dizziness • fatigue • generalized weakness • weight loss
Medication: —
Clinical Procedure: Colostomy
Specialty: General and Internal Medicine

Objective: Unusual clinical course
Background: A 70-year-old African American man presented with fatigue, dizziness, generalized weakness, and considerable weight loss of over 20 pounds in 3 weeks. History-taking revealed he was positive for HIV, hepatitis C, and severe chronic condyloma acuminatum, which had been progressing for 16 years. Treatment and surgical intervention had been continuously postponed due to the patient's long-standing history of heroin abuse.

Case Report: Physical exam and diagnostics showed evidence of sepsis. He was hypotensive, with lactic acidosis and significant leukocytosis, and had acute-on-chronic kidney disease. Urinalysis was positive for nitrites and leukocyte esterase; therefore, broad-spectrum antibiotics were initiated. Additional sources of sepsis were considered due to persistent leukocytosis despite appropriate antibiotic coverage. An MRI of the pelvis was done to evaluate for necrosis of fistulization from potential internal warts as a source of sepsis. The lesions extended from the inguinal areas bilaterally, covering the medial thighs, lower scrotal wall, and wall junction. It had infiltrated the perineum and the entire rectal area, including the gluteal cleft and anus. The patient was consulted by colorectal surgery, urology, and infectious disease services.

Conclusions: Surgical biopsies found that he had both low- and high-grade squamous intraepithelial neoplasia. There was no evidence of invasive carcinoma, which was a concern given his weight loss. Surgery devised a plan that included a diverting colostomy (allowing the infected anal area to heal), followed by resection of his giant condyloma, and re-anastomosing of the bowels to return him to a normal baseline anatomy. A favorable prognosis was expected.

MeSH Keywords: Buschke-Lowenstein Tumor • Condylomata Acuminata • Human papillomavirus 11 • Human papillomavirus 16 • Human papillomavirus 18 • Human papillomavirus 6

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Background

Giant condyloma acuminatum, also known as a Buschke-Lowenstein tumor (BLT) in its severe form, is a slow growing, cauliflower-like lesion that is sexually transmitted. It is most often caused by the human papilloma virus (HPV) strains 6, 11, 16, and 18. Originally identified in 1925, these genital warts are “soft, raised, fleshy lesions found on the penis, vulva, vaginal wall, cervix, or perianal region” [1]. They are rare, locally invasive of the underlying dermal structures, and often benign. However, malignant transformation to squamous cell carcinoma has been reported in advanced cases, and although “transformation to invasive carcinoma is rare (occurring in <1% of cases)” [2], being immunocompromised greatly increases the risk. Patients are often non-responsive to treatment with radiation and chemotherapy. For these reasons, the definitive treatment is surgical resection with wide margins.

Case Report

We present the case of a 70-year-old African American man with HIV (on HAART therapy), who had an advanced case of BLT. His condition had drastically compromised his quality of life and emotional wellbeing.

The patient-reported history indicated that he had been recently compliant with his HAART treatment, and was being followed by the local veteran’s hospital. He had a long-standing habit of heroin abuse that spanned decades. He also had hepatitis C, hypertension, and gout. He presented to the ED with complaints of generalized weakness, dizziness, and weight loss of about 20 pounds over 3 weeks.

The patient appeared frail and somewhat older than his stated age. A giant condyloma acuminatum was present. The lesions were exceptionally painful to palpation and drained a copious purulent-appearing material that was unbearably malodorous. Visual signs of necrosis were not present. The patient was noted to be septic on admission, with hypotension (blood pressure measuring 85/50, which was fluid responsive eventually returning him to his baseline blood pressure of 126/88), lactic acidosis, and a left-shift leukocytosis of 28.18 k. However, he was afebrile. Additional diagnostic data included blood work, revealing low hemoglobin (6.6 g/dL) and hematocrit (21.6%) levels, a chest x-ray that was negative of 28.18 k. However, he was afebrile. Additional diagnostic data included blood work, revealing low hemoglobin (6.6 g/dL) and hematocrit (21.6%) levels, a chest x-ray that was negative for acute process (Figure 1), and a CT scan of the abdomen/pelvis revealing large perineal condylomas without necrosis (Figures 2, 3). The CT scan also divulged considerable stool impaction (Figure 4).

The patient was initially treated for his genital warts with cryotherapy. However, they reoccurred and he never followed up for additional treatment. Over time these warts grew so extensively and shingles. He stated that he still has occasional nerve pain around his right ear from the shingles.

On examination, the patient appeared frail and somewhat older than his stated age. A giant condyloma acuminatum was present. The lesions were exceptionally painful to palpation and drained a copious purulent-appearing material that was unbearably malodorous. Visual signs of necrosis were not present.

The hospital course included empiric treatment of sepsis with Zosyn (2.25 gm 50 ml, IV, Q6H) and Vancomycin injection (1.5 gm 30 ml, IV, once followed by Vancomycin premix in D5W 1 gm 200 ml, IV, Q24 H), that were initiated in the ER and continued for 3 days pending culture results. Blood cultures returned negative. Cultures of the surrounding condyloma tissues came back positive for Pseudomonas aeruginosa and Providencia stuartii. His urine cultures were positive for E. coli, and his antibiotic regimen was changed to Ciprofloxacin (200 mg IV Q12 hr for 7 days) and Metronidazole (7.5 mg/kg IV Q6H for 7 days), as per culture sensitivities.

The patient was retired and had previously worked for a restoration waterproofing company until 2008. He was in the navy from 1968 to 1972, during which time he contracted hepatitis C.

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that currently, they can clearly be classified as a Buschke-Lowenstein tumor (BLT). Each time he consulted with his physician, he was told that his situation could not be resolved surgically given his heroin addiction. He stated that his “inability to conquer my demons and refrain from using heroin for any substantial amount of time” had led to a vicious cycle of relapse, infection, and inability to surgically treat.

The BLT had grown to such epic proportions that it had engulfed the anus. The only way to identify the anal canal was to follow the continuous drainage of liquid stool. The entirety of the lesion was exquisitely tender to palpation, erythematous, indurated, noxiously malodorous, and drained purulent fluid.

The scrotal lesions measured 6×7 cm on the left and 15×8 cm on the right side of the scrotum, involving the scrotal skin (Figure 5). Both testicles were palpable within the scrotum medially to the right perineal condyloma. The perianal aspect of the lesion measured 4 cm laterally and 7 cm superior to inferior. On the right side of the anal verge, there was local soft-tissue loss, with a defect that had condylomatous tissue inside.
of it. A superficial fistula on the rim of this lesion was opened with cautery, and showed no evidence of pus or infection.

Large stool compaction was evident as a result of the BLT. The patient complained of constant constipation pain, even though he was still passing liquid stool (Figure 6). This was concerning for infection.

On digital rectal examination, the perianal condylomatous lesions felt firm within the soft tissue, extending to the level of the sphincter. There was palpable intra-anal extension of the lesion. The anal external sphincter was not clearly discernible due to the vast infiltration of the condyloma. This had led to the patient’s extreme discomfort and fecal incontinence. Chronic irritation of the anal and genital tissue was due to stagnant stool exposure, creating an unhygienic environment. This undoubtedly encouraged bacterial proliferation and prolonged infection. There was no foley or fecal catheter in place.

A sigmoidoscopy was scheduled to assess the degree of tissue involvement and to obtain biopsy samples. Bowel prep was not needed because we were only assessing local tissue involvement up to the distal rectum. The patient, under anesthesia, had a pediatric flexible colonoscope guided into his sigmoid colon. The distal anal canal appeared thickened, and a biopsy was taken, which showed rectal mucosa with no significant pathological abnormalities. Additionally, 4 biopsies were extracted during the procedure. The left perianal condyloma and left perineal and right perineal biopsies all showed low-grade squamous intraepithelial lesions (condyloma acuminate) (Figure 7). The right perianal condyloma biopsy revealed a high-grade squamous intraepithelial lesion with moderate to severe dysplasia (Figure 8). The margins of this excision showed no evidence of invasive carcinoma.

An MRI of the pelvis was negative for rectal fistulas. However, there were distortions of the tissues noted by the radiologist, involving the scrotum, perineum, and rectal region, both superficially and deep (Figures 9–11). There was no evidence of lymph node involvement or metastases.

After sufficient examination, a plan was established to place a diverting colostomy in an effort to maintain a hygienic environment, allowing the soft-tissue infection surrounding the BLT to heal. Surgery was discussed with the patient and his family. A multidisciplinary team consisting of a colorectal surgeon, plastic surgeon, and urologist are collaborating to coordinate an optimal date for the removal of the BLT. This procedure will include surgical excision of the lesion and possible skin grafting due to the immense resection of tissue. If skin grafting is required, it will be via a split-thickness graft harvested from the patient’s front or outer thigh. Upon healing, the patient will require re-anastomosing of the bowels to return him to a normal baseline anatomy. This long-overdue procedure will enable our patient to finally be free of a 20-year torment. In the interim, he has been discharged to a subacute rehab facility. 

Figure 6. Anal lesion obstructing the release of solid feces, leading to constant leakage of liquid stool.

Figure 7. Histopathology slide demonstrating low-grade intraepithelial squamous hyperplasia of the anus.

Figure 8. Histopathology slide demonstrating high-grade intraepithelial squamous hyperplasia of the anus.
Human papillomavirus (Papillomaviridae) is a circular, naked, double-stranded DNA virus that can produce a number of complications, especially if not treated in a timely manner. HPV infection is limited to the basal cells (keratinocytes) of stratified epithelium, the only tissue in which they replicate [3]. It is by far the most common sexually transmitted disease worldwide, and affects 79 million people in the USA, most of whom are in their late teens and early 20s [4].

There are over 170 different strains of HPV identified. They are generally classified as high- or low-risk depending on their propensity for malignancy. Of those 170 types, only around 40 are transmitted through sexual contact and can infect the skin of the anus and genitalia [5]. About 90% of all cases of genital warts are due to strains 6 and 11 [6]. Although only 10% of people infected with HPV manifest genital warts, being immunocompromised is a significant risk factor (as is the case with our patient). In extremely rare instances, common genital warts can develop into Buschke-Lowenstein tumors. These are the giant, slow growing, locally aggressive, and destructive condyloma acuminatum present in the genitoanal region [7].

BLT’s can be differentiated from ordinary condylomas by the characteristic “pushing” rather than “infiltrating” effect, which tends to compress and displace the underlying tissue [8]. They appear as grotesque, cauliflower-like, fungating, ulcerated masses. On histology, they appear as endophytic growths resembling condyloma (papillae with prominent fibrovascular cores, koilocytosis, and broad pushing base), but with more prominent bulbous expansion into underlying tissue [9]. BLTs are known to compromise the penis, vulva, scrotum, bladder, and perianal or anorectal regions.

Treatment can be broken down into 3 categories, depending on severity and location of the lesions, as well as immune status, comorbidities, and social history. The treatment options are:
topical therapy (e.g., using podophyllin, fluorouracil, or radiotherapy), tumor removal (e.g., by cryotherapy using liquid nitrogen, CO$_2$ laser therapy, electrocautery, or surgical excision), and immunotherapy (e.g., using imiquimod) [10].

When genital warts have reached the stage of BLT, surgical intervention is the most effective form of treatment. Unfortunately, as with our patient, most cases progressing to this advanced stage are due to both lack of follow-up and poor lifestyle choices. It should be noted that there is no cure for the underlying HPV infection, and that future reoccurrence is possible.

Conclusions

Although BLT is a rare manifestation of genital warts, which can be caused by multiple different strands of HPV, it is most frequently seen in immunocompromised patients. If left untreated, the risk for malignant transformation becomes a substantial concern. This locally invasive lesion is physically and emotionally debilitating. Regardless of the size and origin of BLTs, early control of the disease using wide, radical surgical excision provides the best overall survival [11]. Patients with BLT should be regularly screened for reoccurrence of new lesions to ensure early diagnosis and prompt treatment. Screening for BLT consists of a physical examination of anal and oral regions, as well as a biopsy of any existing warts.

In our patient’s case, once the lesion is surgically excised, regular appointments with his general practitioner should be implemented. These follow-up visits should include continuous monitoring for the possible recurrence of the BLT. Potential squamous cell carcinoma development, especially when there is high-grade squamous intraepithelial hyperplasia, as in our patient, should be screened for by biopsying any future genital warts that manifest.

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Conflicts of interest

None.

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