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Initial Treatment Outcome For Kaposi's Sarcoma of the Penis in 16 Years Old HIV-Negative Attended at Muhimbili National Hospital in Dar Es Salaam

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1. Abstract

We describe a case of a penile Kaposi's sarcoma in immunocompetent non HIV negative 16 years old without identifiable risk factor. He presented with penile lesions in which diagnosis was confirmed through tissue histopathology and immunohistochemistry (HHV-8). We treated the patient using combination therapy of ABV with different dosage = doxorubicin, bleomycin and vincristine doxorubicin 30 mg/m2 IV, bleomycin 15 units/m2 IV, and vincristine 1.4 mg/m2 IV, administered every 3 weeks. ABV was preceded by a single dose of BV, followed two weeks later by the ABV combination that is subsequently given every 3 weeks for 8 cycles.

We had a holistic approach of which the treatment team to this rare condition included urologist, pathologist and oncologist at Muhimbili National hospital in Dar es salaam.

2. Introduction

Kaposi's Sarcoma (KS) is a multifocal hemorrhagic sarcoma that occurs primarily on the extremities. When it is limited to the penis is rare and a well reported in as a manifestation of Acquired Immune Deficiency Syndrome (AIDS). KS can be confined to the penis in (HIV)-negative patients and found to be related to the herpes virus infection (HHV-8) [1]. It is necessary for the urologist to appropriately and effectively diagnose and manage these lesions in order to optimize outcomes and minimize morbidity [2]. KS can be acquired and the transmission of HHV-8 can be primarily via the saliva but is also transmitted by sexual contact and blood and can occur in patient who are immunocompetent or immunocompromised at variable age groups in different parts of the world [3].

KS is the most common neoplasm of people living with HIV today and in Sub- Saharan Africa it is among the most common cancers in men in overall. It can occur in any immune compromised person infected with Kaposi Sarcoma- associated herpesvirus (KSHV) or human herpesvirus 8. The group of individuals at risk includes elderly, children in KSHV- endemic areas and transplant recipients [4].

The treatment protocols available covers both HIV negative and positive patients but with different modalities at initiation. In HIV positive the emphasis was on initiation of ART and treatment of opportunistic infections followed by sequential combination chemotherapy as described in a Design Schema for a Risk-Stratified and Response-Adapted Therapeutic Approach to Pediatric Kaposi Sarcoma [5]. In this review the management of Kaposi's sarcoma in young boy is being reviewed and follow up initial outcome to be discussed.

3. Case Review

A 16 years old boy from Kigamboni Dar es salaam form two student presented with history penile swellings for 4 months, preceded by itching at tip of penis and progressed to form rashes involving shaft of penis followed by swelling of variable sizes which later ulcerated with foul smelling discharges. The lesions were initially painless but later on become painful. He was circumcised at the age of three years. No history of discharge per urethra. No history of passing blood urine. No reported history of Lower Urinary Tract Symptoms (LUTS), No history of insect bites to the penis, Denied history of tobacco or passive cigarette smoking.

He didn't have any history of fever, weight loss, or mucosal involvement. He had unprotected sex with a female partner of nearly age group six months before initiation of lesions and had no histo-

ry of Sexually Transmitted infections in the past. The patient didn't have a history of chemotherapy, radiotherapy, HIV infection, dermatologic disorders and disease and medications with immune suppression. In his past medical history, there was no report of any kind of disease or surgery.

On assessment was found a young boy, not pale, not jaundiced, not dyspneic, no finger clubbing. Had palpable, matted and tender bilateral inguinal Lymph nodes but no lower Limb Edema. Normal vital signs (BP: 110/70mmHg, PR: 70b/min, T: 37.1 C, RR 18cycles per min) and essentially normal systemic assessment.

On genital examination was found to have multiple ulcerated masses on the glans penile up to mid shaft, with raised margins, mushroom like (Cauliflower) with urethral meatus involvement, tender, normal skin, normal testis palpable in both hemiscrotum, no lesion noted on other parts of the body (Figure 1&2).

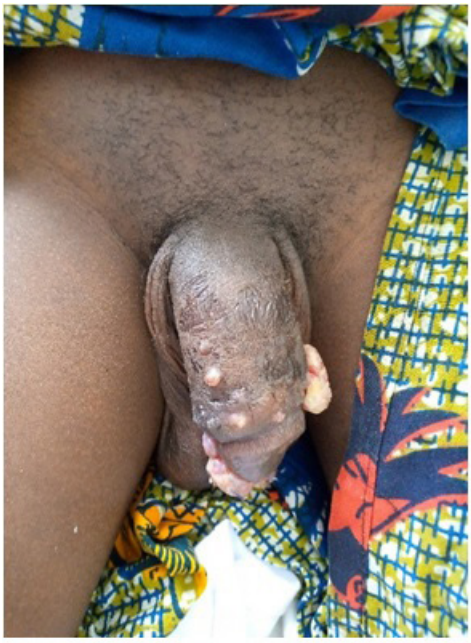

Figure 1

Figure 2

Figure 1 and Figure2: Multiple ulcerated masses on the glans penile

Abdominal pelvic ultrasound and chest X-ray were normal

Laboratory tests done included: Full Blood Count (FBC), VDRL, Treponema agglutination test, urine analysis and urine culture were normal. Serology of HIV, HBV, HCV tests were negative in two times but the Immunohistochemistry HHV8 was detected in tissue by PCR method in biopsy

Wedge biopsy of the lesions had shown tissues formed by nodular lesions with vascular proliferation and marked slits, erythrocytes extravasation and spindle cells intersecting the fibrous stroma (Figure 3&4).

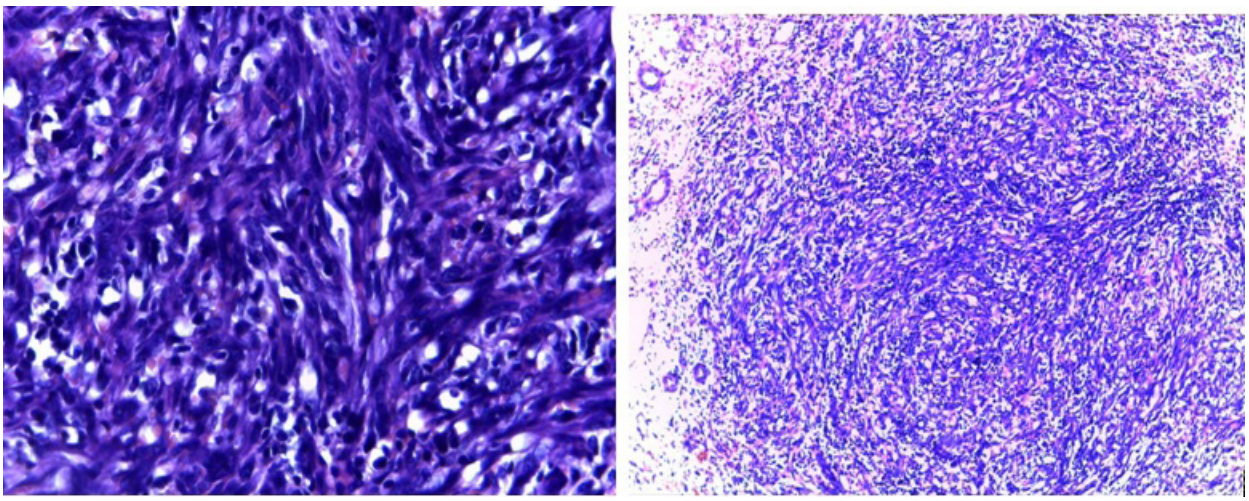

Figure 3 and Figure 4: Tissues formed by nodular lesions with vascular proliferation and marked slits

Modality of treatment given to our patient at Muhimbili National Hospital (MNH) oncology unit was combination chemotherapy. The protocol used was ABV = doxorubicin, bleomycin and vincristine; the dosage includes doxorubicin 30 mg/m2 IV, bleomycin 15 units/m2 IV, and vincristine 1.4 mg/m2 IV, administered every 3 weeks. ABV is preceded by a single dose of BV, followed two weeks later by the ABV combination that is subsequently given every 3 weeks for 8 cycles.

During the treatment the definitions of response to chemotherapy are based upon previously established criteria from the AIDS Clinical Trial Group. Whereas Complete Remission (CR): disappearance of all KS lesions. Very Good Partial Remission (VGPR): a subjective assessment of at least 90% reduction in the size of all lesions and Partial Remission (PR): a subjective assessment of at least 50% reduction in the size of all lesions.

3.1. Follow Up One Month After Two Cycles of Chemotherapy

Lesions subsequently reduced, passing urine normally, no pain or urethral discharge (Figure 5&6).

Follow up two months after completion of 8 cycles of chemotherapy, passing urine normally, no pain, no itching, lesion subsided completely (Figure 7&8) .

Figure 5 and Figure 6: Lesions subsequently reduced

Figure 7 and Figure 8: lesion subsided completely

4. Discussion

We reviewed the management and initial outcome of penile KS with focus on non HIV and young adolescent age group, our case is only 16 yrs old. The occurrence of penile Kaposi's sarcoma in HIV negative is rare and is extremely rare in young's age groups. Being uncommon in HIV negative men, it should be considered as one possibility when treating nonspecific penile lesions [6].

The presentation of KS in a HIV negative individuals needs high index of suspicion and excluding other benign and infectious causes of penile lesion careful pre-test counseling is required and screening for other sexual transmitted infections [7]. Human Herpes Virus 8 (HHV-8) infection is common among children in areas where Kaposi sarcoma is endemic like in our case, however it was well documented that the prevalence of HHV-8 in HIV-positive was twice as high as that in HIV-negative [8].

The diagnosis of Kaposi's sarcoma is based on clinical presentation and histological diagnosis, in our patient the diagnosis was confirmed through tissue diagnosis and immunohistochemistry. The disease grouped currently into four categories of Kaposi sarcoma, including classic, which occurs in patients without known immunodeficiency like to our case which occurs in young men and can be indolent or aggressive; and epidemic or HIV-related, which occurs in patients with AIDS [9].

The benign lesions of penis in immunocompetent patients, even in absence of risk factors for sexually transmitted diseases, should be always investigated, because it could represent the first manifestation of primary KS in which penis could be the only isolated clinical presentation [10] KS limited to the penis is rare and a well-recognized manifestation of Acquired Immune Deficiency Syndrome (AIDS). When KS is confined to the penis is extraordinary in Human Immunodeficiency Virus (HIV)-negative patients [11].

A nearly age group like our case from Somalia a 21-year-old HIV-negative male who no identifiable risk factor on his previous social and family history was diagnosed with an immunohistochemically proven Human Herpes Virus 8 (HHV8)-positive primary penile Kaposi's Sarcoma (KS). He was treated differently with local surgery and was reported recovery for 18months unlike to the index case from Tanzania [12].

Due to its occurrences there is discrepancy in management options in HIV negative patients and there are few literatures however Kaposi's sarcoma in HIV-associated due to the availability of Antiretroviral Therapy (ART) has improved KS survival [13].

The available modalities of treatment for primary penile KS includes local surgical excision, radiotherapy, chemotherapy and laser therapy. The surgical excision is recommended for the small solitary lesions and the radiotherapy is recommended by several authors for large lesions after biopsy and histological confirmation while systemic chemotherapy is used for the severe forms and has shown high success rate [1].

In our setup there is no standardized protocol for management of patient with KS in HIV negative group and they were treated with similar chemotherapy agent but different initiation protocols to those who are HIV positive. In HIV positive group guidelines from different parts of the world recommends initiation of ART and treatment of opportunistic infections especially in newly diagnosed followed by Induction phase chemotherapy then consolidation phase [14].

The chemotherapeutic agent can be monotherapy or in combination, in this case review the combination therapy ABV was used and our patient had very Good Partial Remission (VGPR) in which more than 90% remission was noted on completion chemotherapeutic agent. It has been reported that the overall survival, event-free survival, and quality of life appear to have improved with three-agent combination chemotherapy as compared to monotherapy agent as in our case review [15].

In Southern West Tanzania it was found that the use of monotherapy in KS relapse and those with contraindication to bleomycin, vincristine, and doxorubicin (ABV), had significant response on Paclitaxel on clinician preference thus high rates of long-term survival and favorable outcomes were possible with paclitaxel treatment but most patient are HIV positive. In further review of use of paclitaxel on HIV negative endemic KS it was highlighted that it has important therapeutic role in the management of pediatric patients with endemic KS [16].

Depending on the nature of lesion and extent of disease treatment decision can varies from minimal invasive to invasive, the use of immune modulator was suggested for superficial penile KS. A reported a case of Kaposi's Sarcoma (KS) in an HIV-negative man that was successfully treated by topical application of imiquimod 5% cream (Aldara) after failure of using cryotherapy [17].

The outcome of treatment modalities offered is variable depending on extent of disease and identified risk factor, immune status, age at diagnosis. In general, Unlike KS in AIDS patients, non-AIDS associated KS is a rather localized process which rarely involves lymph nodes in most cases are responsive on local or systemic therapeutic strategies [18].

Since the incidence of a cancer diagnosis in children and young adolescents is increasing. Impact on fertility potential should be discussed to patient's prior initiation of chemotherapy or other modalities of treatment for KS. Our patient received chemotherapy and is at 16 years old, since the impact on reproductive hormones could not be known at completion of chemotherapy cycle, thus needful long term follows up highly recommended [19]. Risk of infertility varies substantially with patient and treatment modalities and there are associated factors affecting future fertility in this group of patients [20].

5. Conclusion

KS is uncommon in young and very rare in young HIV negative

group and there should be high index of suspicion when lesions of penis in immunocompetent patients is noted, even in absence of risk factors for sexually transmitted diseases and should be investigated as it could represent the initial presentation of primary KS in which penis could be the only isolated in clinical presentation. The surgical management could represent a good therapeutic option in localized lesion and chemotherapy recommended in systemic disease leading to disease clinical resolution. It is also important to consider quality of life and sexual functions as one of the important aspect especially in young age group who are sexually active.

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