

Squamous cell carcinoma of penis with bullous pemphigoid masquerading as lymphogranuloma venereum

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Abstract

We report a case of a 60-year old unmarried male who presented with multiple ulcers and foul smelling discharge from the groin since 4 months and multiple tense bullae over the trunk of 1 month duration. Groove sign was present. Investigations for lymphogranuloma venereum (LGV) and other sexually transmitted diseases were negative. Histopathology from the ulcer in the groin and growth in the penis revealed squamous cell carcinoma (SCC). Skin biopsy of bulla was diagnostic of bullous pemphigoid (BP). We report a rare case of SCC masquerading as LGV with BP occurring as a paraneoplastic phenomenon.

Key words: Bullous pemphigoid, paraneoplastic phenomenon, squamous cell carcinoma of penis

INTRODUCTION

Penile cancer is rare in developed countries, whereas in India and other Asian countries the incidence is decreasing. The incidence of squamous cell carcinoma (SCC) of penis varies worldwide with age, circumcision, and hygiene practices. Phimosis, lack of neonatal circumcision, human papillomavirus (HPV) infection, penile lichen sclerosis, exposure to tobacco products, and Psoralen ultraviolet A (PUVA) are the known risk factors for development of SCC of penis. Up to 50% of patients with SCC of penis delay seeking medical attention by more than a year due to embarrassment, guilt, fear, ignorance, and personal neglect. The level of

denial is substantial, given that the penis is observed and handled on a daily basis.

CASE REPORT

A 60-year-old unmarried male, presented with a history of multiple ulcers over his right groin of 4 months duration. It started as a painless swelling over the right groin, which evolved into multiple ulcers with foul swelling discharge. One month later, he developed a similar swelling in the left groin. There was no history of trauma at the site. Patient denied history of sexual exposure and there was no history of genital ulcer. He does not give history of arthralgia, loss of weight, or loss of appetite. Bowel and bladder habits were normal.

The patient also gave history of multiple blisters over the trunk and extremities of 1 month duration. There was no history of oral erosions or drug intake prior to the onset of lesions.

On general examination, the patient was moderately built and nourished, there was no pallor, icterus,

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pedal edema, or generalized lymphadenopathy. His systemic examination was normal.

Examination of the right inguinal region showed three tender excavating ulcers of sizes 6 × 8, 5 × 7, and 3 × 5 cm, respectively, located adjacent to each other with rolled out, everted edges, and foul smelling purulent, blood-stained discharge. Enlarged, grouped, and matted tender inguinal lymph nodes of sizes 5 × 7 cm were felt below the ulcer. Examination of the left inguinal region also revealed enlarged, grouped, tender inguinal nodes of size 8 × 6 cm present both above and below the inguinal ligament giving the appearance of “Groove sign of Greenblatt” [Figure 1]. External iliac lymph nodes were not palpable.

Genital examination revealed phimosis. A firm, indurated, non tender mass of size 3 × 2 cm was felt through the prepuce skin on the glans penis, located on the 11’o-clock to 2’ o-clock position. There was no lymph edema of penis. Per-rectal examination was normal.

Multiple tense bullae filled with clear fluid and erosions were present over upper limbs, lower limbs [Figure 1], and trunk. A tense bulla was present over the shaft of penis, which eroded later. Nikolsky’s sign and bulla spreading sign were negative. There were no oral erosions.

With the above clinical findings, a differential diagnosis of lymphogranuloma venereum (LGV) or SCC of penis with regional metastasis associated with bullous pemphigoid (BP) occurring as a para neoplastic phenomenon were considered.

Dark field smear was negative for *Treponema pallidum*. Tissue smear was negative for Donovan

bodies. Gram’s stain was negative for gonococci. IgG antibody for *Chlamydia trachomatis* (serovars L1, L2, L3) was negative. VDRL for syphilis and ELISA for human immunodeficiency virus (HIV) 1 and 2 were nonreactive. Mantoux was also negative. No abnormality was seen in his urine microscopy and pus culture reports. His laboratory investigations revealed anemia. Other blood counts, liver and renal function tests were normal. Tzanck smear taken from the bulla did not show acantholytic cells. His chest x-ray and ultra sound of the abdomen were normal.

Wedge biopsy of the ulcer in the right groin and from the penile growth showed ulcerated hyperplastic epidermis with severe dysplasia. The dysplastic epithelial cells invade the basement membrane forming nests with keratin pearls, consistent with keratinizing SCC of penis [Figure 2]. Skin biopsy taken from a tense bulla over the trunk, showed a sub epidermal bulla with mild lymphocytic infiltrate in the upper dermis consistent with BP.

Hence, a diagnosis of keratinizing SCC of penis with regional lymphnode metastasis and BP was confirmed. Patient was referred for surgical opinion and management.

DISCUSSION

SCC of penis may present either as a flat growth, infiltrating, or papillary growth. The lesion is not visible in most patients since the prepuce is non-retractile. The lesion continues to grow, with spread to inguinal nodes. The inguinal nodes increase in size followed by erosion of the skin over the nodes. Death may result from erosion of the femoral artery.^[1,2]



Figure 1: Right groin showing multiple excavating ulcers, left groin shows ‘Groove sign of Greenblatt’ and multiple tense bullae with erosions over thighs and penile shaft

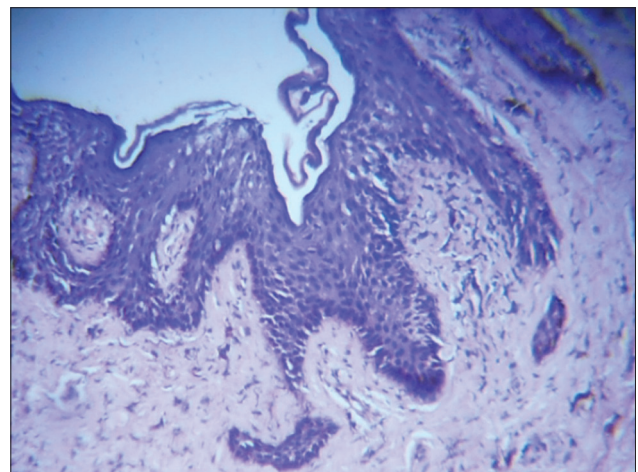


Figure 2: H and E section of penile growth shows ulcerated hyperplastic epidermis with severe dysplasia. The dysplastic epithelial cells are seen to invade the basement membrane

LGV primarily affects the lymphatic system. It starts as a small transient, inconspicuous lesion on the genitalia and spreads to regional lymph nodes resulting in bubo and suppurative lymphadenopathy.^[3,4]

In our patient, SCC of penis was masquerading as LGV since he presented with history of inguinal bubo, which later ulcerated. In addition the “Groove sign of Greenblatt” was positive. Moreover, the primary SCC was not noticed by the patient and was not visible during examination since the prepuce was non-retractile. A biopsy from the groin ulcer and penile growth that was felt on palpation had confirmed the diagnosis.

The occurrence of BP in this patient can be attributed as a paraneoplastic phenomenon to SCC of penis. The association between BP and malignant diseases remains controversial. Pemphigoid has been reported in numerous patients with malignancies, like breast cancer,^[5] B-cell lymphoma,^[6] lung cancer, gastric cancer, cancers of colon or rectum, endometrial cancer, and many others.^[7,8] In SCC, the expression of BPA1 and BPA2 is clearly increased.^[9,10] This could lead to production of antibodies directed against these antigens that results in the development of BP in these patients. To the best of our knowledge this is the first case of SCC of penis with BP occurring as a paraneoplastic phenomenon. We present this case to highlight that SCC of penis can masquerade as LGV and a high index of suspicion is required for an early diagnosis of SCC penis.

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