A rare presentation of pseudoepitheliomatous keratotic and micaceous balanitis with malignant transformation

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Abstract

Pseudoepitheliomatous keratotic and micaceous balanitis (PKMB) is a rare nonvenereal penile condition with a risk of malignant transformation. We report a case of PKMB in a 45-year-old male who presented with multiple keratotic growths over his glans penis after circumcision for long-standing phimosis. Histopathology revealed pseudoepitheliomatous hyperplasia and atypical cells. He was successfully treated with wide local excision.

Key words: Balanitis, micaceous, pseudoepitheliomatous

Introduction

Lesions on the penis can be a challenge to diagnose often due to late presentation, self-medication, or prior ineffectual treatment causing alarm to the patient. Certain rare conditions of the penis can progress into malignant growths requiring invasive surgery. One such exceedingly rare premalignant lesion is pseudoepitheliomatous keratotic and micaceous balanitis (PKMB) which is outlined by mica-like scales and keratotic horny masses over the glans penis.[1] Although the etiology of PKMB is perplexing, it is generally seen in the elderly male population. It has a potential to evolve into squamous cell carcinoma and verrucous carcinoma rarely.[2] Herein, we report a case of PKMB presenting as multiple cutaneous horny growths in an adult male.

Case Report

A 45-year-old male presented with asymptomatic penile growths of 8-month duration. He first noticed it as a small elevated keratotic growth post circumcision, which was done for preexisting phimosis 10 months back. The lesions had been gradually increasing in size and number. There was no history of preceding trauma or any systemic disease. He gave no history of exposure to the risk of sexually transmitted disease. Prior treatment included the use of topical antibiotic cream without any improvement in symptoms. Clinical examination showed multiple bark-like, hard, yellowish horny masses over the glans penis sparing the urethral meatus [Figure 1]. Lesions were confined to the glans penis, whereas examination of the penile shaft and scrotum was normal. Inguinal lymph nodes were not palpable. The patient’s serology was negative for HIV, syphilis, and hepatitis B and C viruses. Routine laboratory investigations which comprised basic blood counts, levels of blood glucose, and tests for renal and liver function showed normal values. Based on these findings, excisional biopsy of the lesion was performed heeding toward a diagnosis of PKMB. Histopathology of the lesion revealed pseudoepitheliomatous hyperplasia with extensive hyperkeratosis, parakeratosis, acanthosis, individual cell keratinization, and atypical cells. Dermis showed dense lymphoplasmacytic inflammatory infiltrate [Figure 2a and b]. A final diagnosis of PKMB evolving to squamous cell carcinoma of the glans penis was made, and the lesions were surgically excised with wide margins.

Discussion

In 1961, Lortat-Jacob and Civatte first described a case of PKMB characterized by thick scaly plaques on the glans penis. “Micaceous” literally means mica-like, resembling sheets of white scaly aluminum silicate. It was initially considered an entirely benign condition.[3]
At present, it is reflected upon as a premalignant condition with low-grade malignancy potential. Pseudoepitheliomatous response to chronic inflammation has been proposed as a possible etiopathogenic mechanism.[4] Kang et al. reported a case of recurrent PKMB associated with human papillomavirus and postulated the role of transformation to verrucous carcinoma.[5] Acquired phimosis and adult circumcision have often been linked with this condition.[6] Clinically, PKMB presents as an asymptomatic slow-growing scaly or verrucous plaques on the surface of the glans penis, rarely causing urinary obstruction. The pathogenic evolution of PKMB involves four stages: (a) plaque stage (early), (b) tumor stage (late), (c) verrucous carcinoma, and (d) squamous cell carcinoma with lymphatic and metastatic invasion.[7] Histologically, initial stages show marked hyperkeratosis and parakeratosis, acanthosis, and pseudoepitheliomatous hyperplasia. In later stages, features of cellular atypia, koilocytes, and pleomorphism are seen.[6] Differential diagnoses include lichen sclerosus et atrophicus, penile psoriasis, penile horn, giant condyloma, verrucous or squamous cell carcinoma, and erythroplasia of Queyrat. Treatment with topical 5-fluorouracil, radiotherapy, cryotherapy, and CO₂ laser has been used when there is no histological evidence of malignancy. Topical 5-fluorouracil is the most highly recommended treatment for the initial plaque stage.[8,9] Long-term surveillance with posttreatment biopsies is recommended during the follow-up period. The tumor stage of PKMB showing features of dysplasia and invasion is defiant to topical treatment. Wide local surgical excision, resurfacing and glansectomy with split-thickness skin graft are recommended depending on the progression of the lesion in advanced cases having histological features of cellular atypia.[10]

Our patient presented at a relatively young age with preexisting long-standing phimosis. Adult circumcision could be a possible trigger for PKMB in our patient. PKMB being a rarity should be recognized early due to its association with penile malignancy. Early clinical as well as histopathological identification and prompt treatment is undoubtedly the best course of action for the management of this condition. The basic understanding of this limitative ailment could be better inferred with a lifelong follow-up of the patient.

**Declaration of patient consent**
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**
There are no conflicts of interest.

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