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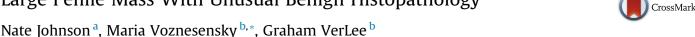
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Large Penile Mass With Unusual Benign Histopathology



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ABSTRACT

Pseudoepitheliomatous hyperplasia is an extremely rare condition presenting as a lesion on the glans penis in older men. Physical exam without biopsy cannot differentiate malignant from nonmalignant growth. We report a case of large penile mass in an elderly male with a history of lichen sclerosis, highly suspicious for malignancy. Subsequent surgical removal and biopsy demonstrated pseudoepitheliomatous hyperplasia, an unusual benign histopathologic diagnosis with unclear prognosis. We review the literature and discuss options for treatment and surveillance.

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Introduction

Pseudoepitheliomatous hyperplasia is a rare penile pathology considered a distinct clinical entity by some, while others view it as a subset of verrucous carcinoma with premalignant potential.^{1,2} Given the lack of data and sparse literature regarding this unusual entity, pseudoepitheliomatous hyperplasia continues to pose a challenging clinical problem with uncertain prognosis.³

Case report

A 73-year-old male with a history of penile lichen sclerosis and chronic leukemia presented with a growth on his penis. The mass was not symptomati, but was causing occasional dark spotting on his underwear. He described no urinary symptoms and had no other complaints.

Physical examination revealed a mass measuring 1.5×2.5 cm, attached to the ventral aspect of the glans penis (Figs. 1 and 2). The mass was pedunculated, nodular, friable, and noted to be weeping dark serosanguinous fluid. Given the high concern for malignancy, the patient was scheduled for surgical excision, and consented for possible partial penectomy pending pathology results.

Pre-operative staging contrast-enhanced computed tomography of the abdomen and pelvis revealed bilateral but non-specific inguinal, iliac and retroperitoneal lymphadenopathy, suspicious for malignancy.



Figure 1. Right lateral view.

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Figure 2. Anterior view.

The mass was widely excised, and intra-operative pathologic frozen sections yielded a diagnosis of myxoma, without evidence of tumor at the surgical margin. No penectomy was performed.

Subsequent pathology analysis demonstrated a benign atypical squamoproliferative and spindle cell lesion with clear margins (Fig. 3). The tissue sample was felt to represent an extremely rare form of benign pseudoepitheliomatous hyperplasia.

A follow up CT scan 4 weeks later demonstrate stable pelvic and retroperitoneal lymphadenopathy compared to original CT. In the setting of his history of chronic leukemia these were felt to be stable and the patient was referred to his hematologist for further follow-up. The patient will be closely observed with biannual physical exams.

Discussion

Penile pseudoepitheliomatous hyperplasia is a rare histopathologic diagnosis affecting the glans penis, first described in 1961 as pseudoepitheliomatous keratotic and micaceous balanitis (PKMB) of Civatte. It characteristically presents as a white, keratotic plaque on the glans in the elderly. While little is known about the risk factors or pathogenesis of PKMB, this disease appears to be most closely associated with adult circumcision. In this patient, neither of these typical findings or associations exists. However, this patient had a history of penile lichen sclerosis, similar to a case of PKMB reported by Bashir et al in 2010. While penile lichen sclerosis is associated with up to 9% risk of subsequent squamous cell carcinoma, to date there is no recognized association between penile lichen sclerosis and PKMB.

Regarding prognosis, a review of the literature by Perry et al in 2008 revealed a total of 14 case reports of PKMB. Five patients progressed to verrucous carcinoma, four progressed to squamous

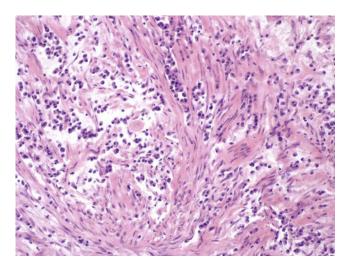


Figure 3. Tissue histopathology demonstrating spindle cells.

cell carcinoma, and one patient progressed to verrucous carcinoma and then subsequently to squamous cell carcinoma. The authors concluded that PKMB has malignant potential if inadequately treated.⁵

Our patient had a successful excision of the mass with negative surgical margins. However, his prognosis and risk of recurrence or progression to malignancy is uncertain. This may be especially true given that the mass in our patient was atypical for PKMB as it was a large and pedunculated, rather than the more characteristic plaque. Ultimately, our patient has been adequately treated with surgical excision and we will continue to monitor him for clinical recurrence.

Conflict of interest

The authors declare they have no conflicts of interest.

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