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Non-cutaneous presentation of mycosis fungoides involving the penile corpora: Case report and review of current literature

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A B S T R A C T

Mycosis fungoides involvement of genitalia is rare. We present a 63-year-old man with history of cutaneous T cell lymphoma with large cell transformation status post multiple electron beam radiation cycles who presented with a new, enlarging penile mass. He underwent ultrasound, MRI, and excisional biopsy. Pathological results indicated hematogenous spread of T cell lymphoma with large cell transformation. Peri-operative radiation was performed, and the patient had significant reduction in penile mass size but some subsequent erectile dysfunction. In discussion of this case, we examine management of penile mycosis fungoides.

1. Introduction

Mycosis fungoides (MF) is a mature T cell non-Hodgkin lymphoma which commonly presents as slowly progressive, variable skin lesions. While any skin surface may be affected, the most common presentation of MF typically arises on the "bathing trunk distribution." More advanced stages of MF include involvement of lymph nodes, blood, and other viscera, commonly the spleen, liver, and GI tract. While all systems may be affected by MF, the genitalia are rarely involved.[1]

The pathophysiology of MF includes the dysfunction of T-cells which subsequently leads to the suppression of host cellular immunity.[2] Patients with advanced MF are at an increased risk of infection and other secondary malignancies, therefore treatment is imperative. Treatment of limited disease involves local radiation and/or topical agents, while more extensive disease involves treatment with systemic agents or hematopoietic cell transplantation.[3] Surgical excision may also be necessary if increased tumor burden impacts quality of life. Herein, we report a case of Stage IIb mycosis fungoides with involvement of penile corpora treated with peri-operative local electron beam radiation.

2. Case description

A 63-year-old circumcised male with history of Stage IIb mycosis fungoides (MF) subtype of cutaneous T cell lymphoma (CTCL) with large cell transformation status post palliative electron beam radiation to multiple solid cutaneous lesions presented for evaluation of new enlarging subcutaneous penile mass. He reported superficial pain with erections. He denied penile pain at rest, erectile dysfunction, penile curvature, dysuria, or other lower urinary tract symptoms (LUTS). On physical exam, a firm, nontender, well-demarcated, mostly circumferential penile lesion was palpated extending from the penopubic angle at the mid-shaft approximately 5 cm in length and extending from the 12 o'clock position dorsally to the 9 o'clock position on the lateral shaft. Corporal involvement by physical exam was unclear. The urethra was unable to be clearly palpated as the lesion appeared to overly the urethra ventrally. Some palpable small, mobile, nontender bilateral inguinal lymph nodes were also noted. A penile ultrasound was performed which showed a soft tissue mass thought to involve cutaneous tissue of the left mid shaft. The lesion did not appear to involve the corpora on ultrasound (Fig. 1). However, MRI was obtained which did show a 3.7 × 5.7 × 3.9 cm enhancing mass involving the left corpora (Fig. 2).

He then underwent excisional biopsy and diagnostic cystourethroscopy. There was no evidence of urethral involvement, and two wedge biopsies were taken. The immediate post-operative course was uneventful. Biopsy confirmed CTCL with variably positive CD3 and CD4 (Fig. 3). He was treated with peri-operative local penile radiation (12Gy in 4 fractions). He reported a significant size reduction in penile mass following therapy. At five-week follow up, physical exam demonstrated significant size reduction of the large subcutaneous penile mass with mild edema and a well healing distal ventral penile incision with minimal tenderness and no evidence of infection or drainage. He did endorse some erectile dysfunction and decreased penile sensation.

Abbreviations: MF, Mycosis Fungoides; CTCL, Cutaneous T-Cell Lymphoma.

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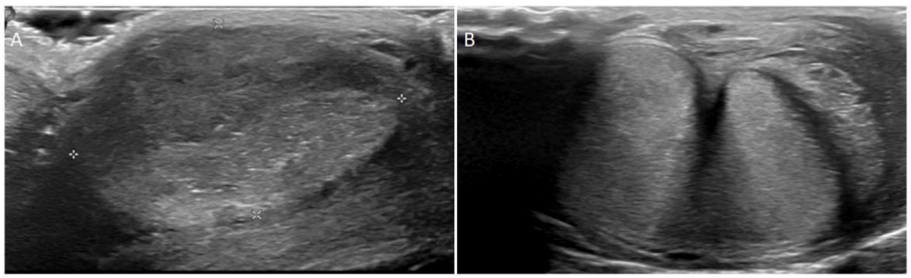

Fig. 1. A) Penile Ultrasound, long view, showing a nonspecific well-defined penile mass; B) Penile Ultrasound, transvers view, which does not appear to show any concrete corporal invasion.

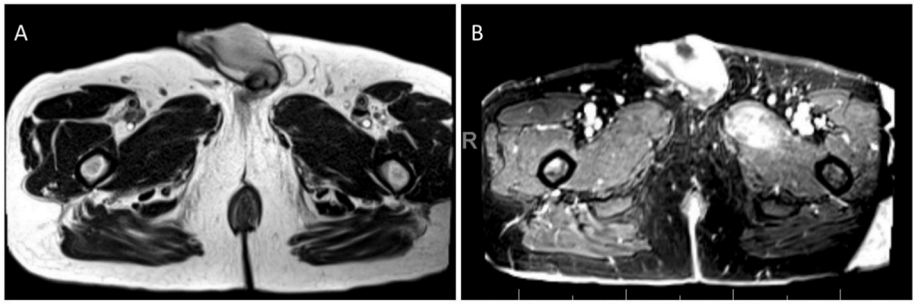

Fig. 2. A) MRI Pelvis showing a 3.7 × 5.7 × 3.9cm moderately T2 hyperintense lesion which involves the left corpus cavernosum in the mid penile shaft; B) Lesion demonstrates enhancement with an area of central necrosis.

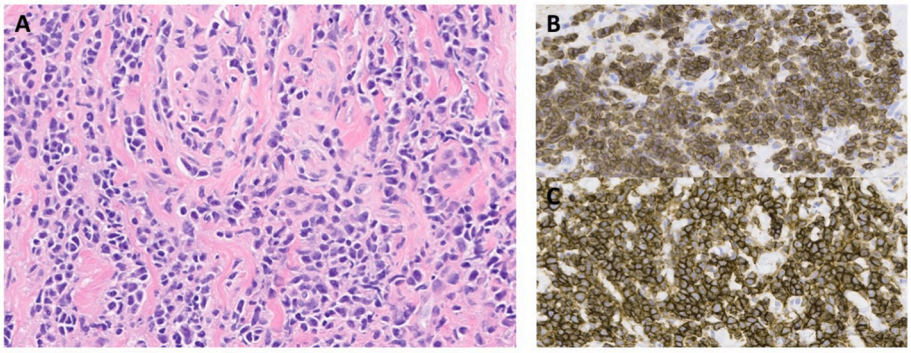

Fig. 3. H&E section of penis biopsy shows diffuse abnormal lymphoid infiltrate within the smooth muscles. The lymphocytes are medium to large sized with irregular nuclei, one to several nucleoli, and clear cytoplasm. No necrosis noted (A). The atypical lymphocytes are negative for CD7 and CD8 and are positive for CD3 (B) and CD4 (C).

3. Discussion

MF with genital involvement has been previously described, however it is very rare. Prior cases have described treatment of penile shaft skin lesions with a combination of multimodal treatments, including topical therapies, radiation, and systemic therapies.[4] While penile MF has been previously reported, our case is unique in that the lesion involved the corpora cavernosa rather than the penile shaft skin. Given the ample vascularization of the penis, we postulate that this represents hematogenous spread of disease as may be seen with other metastatic malignancies.

Metastases to the penis can develop from arterial, venous, and lymphatic mechanisms, as well as through local invasion. Mearini et al. examined around 500 cases of penile metastases reported in the literature and found the most common primary sites to be bladder, prostate, rectosigmoid, and kidney, followed by lymphoma as the fifth most common primary cancer.[5] As the lesion in our case invaded the corpora, topical therapy would be rendered ineffective and thus our patient received radiation. The amount of radiation used in our treatment of 12Gy in 4 fractions to the penile mass is the lowest reported dose to date. Perioperative local radiation offers a relatively rapid response to skin lesions to prevent progression of disease and clinical appearance with few side effects. Repeated treatments with local radiation may be offered for new penile lesions. Radiation to the penis may result in unique complications such as erectile dysfunction, urethral strictures, and meatal stenosis, so patient counseling is critical to optimize quality of life and disease-free outcomes.

Though radiotherapy was successful in treating this case of penile MF, other cases have been treated with systemic therapy. Schaufler et al. described a case of a recurrent stage IB MF who presented with new-onset penile lesions and lower urinary tract symptoms refractory to narrow-based ultraviolet therapy, methotrexate, and imiquimod.[6] His disease progressively involved other systems, and systemic brentuximab (1.8mg/kg every three weeks) was initiated. His penile lesions and symptoms responded well to systemic therapy. Importantly, this is the first described case treated with systemic therapy. In our case, systemic

therapy was considered given recurrent disease, but because our patient's lesions had responded to radiation in the past the patient elected for a repeat course of radiation. In the future, systemic therapy should be considered for patients with penile MF, especially in cases of suspected hematogenous spread or if concerned about local side effects of radiation.

4. Conclusion

MF often affects the truncal skin, but involvement of other locations including the genitalia is rare. We demonstrate decreased disease burden of a corporal penile MF with large cell transformation after diagnostic wedge biopsy and local radiation alone. Post-treatment, our patient reports interval erectile dysfunction and decreased penile sensation 1 month following penile radiation. Corporal involvement likely represents hematogenous disease spread, and radiation to the penile tissue may have unique complications affecting the genitals which may not be considered with radiation to other areas of the body. Further investigation in MF management must be explored to determine the optimal treatment to maximize disease response and limit urological complications.

5. Contributions

(I) Conception and design: All authors; (II) Administrative support: S. Krzastek; (III) Provision of study materials or patients: None; (IV) Collection and assembly of data: None; (V) Data analysis and interpretation: None; (VI) Manuscript writing: All authors; (VII) Final approval of manuscript: All authors.

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Declarations of competing interest

None.

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