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Case Report

Primary Malignant Melanoma of Male Urethra: A Rare Occurrence

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Abstract

Introduction: Mucosal melanoma is a subtype of Malignant Melanoma (MM). Urethral malignant melanoma is an extremely rare entity with poor prognosis. It is associated with early recurrence and metastases.

Methodoly: It is a retrospective case report. Data was retrieved from Human Information System (HIS), an electronic data-based system of Shaukat Khanum Memorial Cancer Hospital and Research Centre.

Case Discussion: We presented a case of MM of urethra. Forty-year-old male presented with hematuria and mass in penile urethra for 6 months. Cystoscopy and excision of mass was performed which showed malignant melanoma. After complete pre-operative workup and multi-disciplinary team meeting, anterior urethrectomy and suprapubic cystostomy was performed. Adjuvant interferon was given. However, patient presented with bilateral inguinal lymph node metastases which was detected on Positron Emission Tomography scan. He was offered radical treatment. He did not consent for the surgery. Patient died after 1 year of completion of treatment.

Conclusion: With little knowledge reported, the management of urethral MM is still controversial. Proper planning and aggressive nature of treatment might help in curative management.

Keywords: Malignant Melanoma; Mucosal Malignant Melanoma; Genitourinary Malignant Melanoma; Rare Urethral Cancer; Malignant Melanoma of Urethra

Introduction

Malignant Melanoma (MM) can affect any part of the body. There are two types of malignant melanoma i.e. cutaneous MM and mucosal MM. Primary mucosal MM is a rare occurrence which constitutes 4% of all MM [1]. Mucosal MM can arise from all mucosal surfaces of the body, however it mostly arises from head and neck, female genital tract, anorectal region, and urinary tract [2]. Melanocytes arises from neuroectodermal cells and urethra arises from endoderm, hence the incidence of malignant melanoma of urethra is rare [1,3].

First case of urethral MM was reported by Reed [4]. After that, some case reports and series have been published [1,5]. The largest pool of data was published El-Safadi et al in 2013 [6]. The disease is more predominant in females. The clinical presentation of urethral MM is similar to that of urothelial carcinoma, resulting in a delay of diagnosis [7]. Despite aggressive treatment of disease, prognosis of disease still remains poor [8].

We reviewed the data from 1st January 2000 till 31st December 2019. 271 patients with a primary diagnosis of MM were registered at Shaukat Khanum Memorial Cancer Hospital and Research Centre. 174 patients had primary cutaneous MM while 97 patients had primary mucosal MM. Only one patient had MM of urethra which constitutes 0.36% of all MM and 1% of all mucosal MM. In this case report, we present a case report of MM arising from urethra and discuss the clinical management of disease.

Case Report

Forty-six years old presented in outpatient department with history of hematuria and mass palpable in the last 6 months. He had addiction of smoking 20 packs of cigarettes. He had no comorbidities and denied any family history of cancer. Abdominal examination was unremarkable. Cystoscopy and excision of mass was performed which showed mass in penile urethra. Histopathology slides showed nests and sheets of malignant neoplasm composed of spindle shaped to epithelioid cells. There is moderate cytological atypia with pleomorphic vesicular nuclei and prominent nucleoli. Intracytoplasmic melanin is also evidence. There is brisk mitotic activity as well as areas of necrosis. Immunohistochemistry showed positivity for HBM45 and melan A thus confirming malignant melanoma. After the surgery, metastatic workup was performed which was negative.

Urethrogram was performed which showed mucosal irregularity near navicular fossa along with filling defect in bulbous urethra hinting towards the skip lesion of the known urethral malignant melanoma. Positron Emission Tomography scan (PET) confirmed urethrogram findings. However, no FDG avid metastatic activity was observed. Magnetic Resonance Imaging (MRI) scan showed soft tissue mass which is involving the vulvar part of the anterior urethra, with expansion and invasion into the corpus spongiosum, but membranous urethra and pelvic diaphragm had normal appearance. Patient was discussed in Multi-Disciplinary Team (MDT) meeting;

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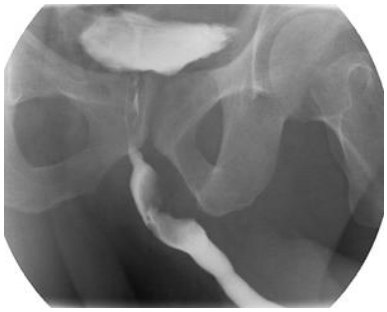

Figure 1A: Urethrogram showing skip lesions.

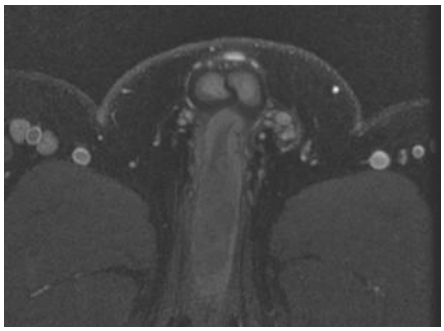

Figure 1B: Mass in the anterior urethra on MRI scan.

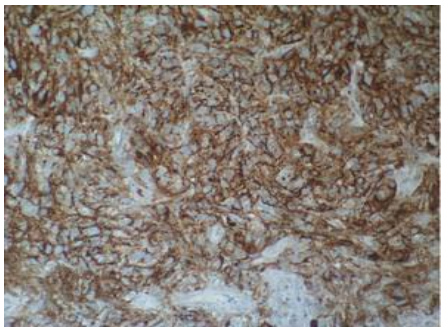

Figure 1C: HMB-45 expression in melanoma cells.

the recommendation from MDT was to perform surgical resection followed by adjuvant interferon therapy.

Anterior urethrectomy and suprapubic cystostomy was performed. Operative finding was palpable tumor in bulbar urethra. Histopathology was consistent with mucosal MM and all of the margins were uninvolved with MM. The stage of tumor was pT1. Patient remained stable during his post-operative period and was discharged on 4th day of surgery. Patient was given adjuvant doses of interferon for 3 months. Then he was kept on surveillance.

On his first visit after completion of therapy, bilateral inguinal lymphadenopathy was appreciated on clinical examination. PET scan showed bilateral FDG avid inguinal lymphadenopathy. Ultrasound guided Fine Needle Aspiration (FNA) confirmed the metastatic MM of inguinal lymph nodes. After discussion in MDT, patient was offered the option of radical cystoprostatectomy with bilateral inguinal lymph node dissection. He refused for further management

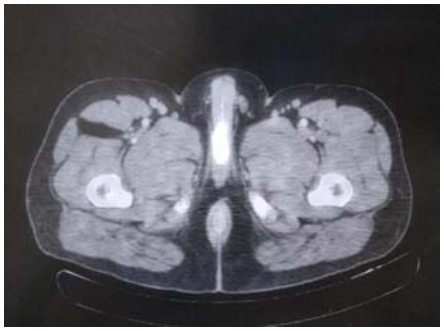

Figure 1D: PET scan showing mass of anterior urethra with inguinal lymph nodes.

and wanted to try herbal medications. Unfortunately, he died after 1 year of surgical treatment due to metastases of disease.

Discussion

Cutaneous MM is the commonest subtype of MM, comprising of 81 of all cases of MM. Involvement of male urethra by MM is an extremely rare entity. Urethral MM accounts for 0.2% of all MM while 4% of all urethral MM (9-11) which can be seen in our reported case. Distal urethra is most common site involved by MM [6] as in our case report distal urethra was involved.

Our patient presented earlier than the reported mean age of diagnosis of mucosal MM [1]. Male patients present slightly earlier than female patients [6]. Most common symptoms are urethral mass, dysuria, local bleeding, hematuria, incontinence, vaginal bleeding, non-specific perineal pain, vaginal discharge and weight loss [6,12]. Our patient presented with hematuria and urethral mass for 6 months. Mean duration of symptoms reported is 21 weeks, however this patient presented late. Urethral MM are mistaken for either benign lesion including mucosal prolapse and chancre or malignant lesions [12,13].

The pathogenesis of urethral malignant melanoma is still unknown and several theories have been proposed. A possible mechanism is metaplastic change in the squamous and glandular epithelium into pigment producing cells (Melanocytes) [8]. Immunohistochemical markers are used in the diagnosis of MM as on the microscopic examination, there is wide histological spectrum of patterns. Most common used markers are S-100 protein and HMB-45. In our case report, we used HMB-45 for confirming the histological diagnosis of MM. HMB-45 is considered quite specific for melanocytic neoplasm [14].

Majority of the times, patients present with metastases due to aggressive biology of the disease. Despite of the radical management which involves radical surgery, chemotherapy, immunotherapy and radiotherapy, the prognosis still remains poor [12]. We did the same for the management of our patient; we started with the pre-operative workup to know the extent of disease and the metastases if present. The curative surgery was performed with achievement of R0 resection of tumor. Inguinal lymph node dissection is still debatable. Some suggests performing lymph node dissection at the time of primary surgery [15]. Inguinal lymph nodes are the first site of metastases in distal urethra MM. Although, we did not perform inguinal

lymph node dissection at the time of primary surgery and then on surveillance, patient presented with inguinal lymphadenopathy; but the evidence from this case report is not enough in taking the decision of inguinal lymph node dissection. The optimal surgical treatment for this disease is still controversial.

Mucosal MM is associated with early recurrence and early metastases as it is seen in our study. Despite surgical treatment and adjuvant therapy, patient presented with early metastases. Metastases to inguinal lymph node occur due to spread through deep lymphatic system. Palpable tumor at presentation is a sign of advanced disease. It is observed in our patient that he presented late after 6 months of symptoms and had palpable mass in the urethra, these two factors indicate towards the aggressive nature of disease resulting in poor prognosis. In our case, we did have the chance of performing more extensive resection and offering further adjuvant chemotherapy or immunotherapy but patient did not consent for it.

Conclusion

MM of distal urethra in male is an extremely rare entity. Despite radical treatment including surgery, chemotherapy, immunotherapy and radiotherapy; the prognosis of the disease is still dismal. This case report will add the information from this part of the world regarding the management of urethral MM.

Consent: Consent was obtained from the patient prior to the management of disease.

Ethical Approval: Due to the retrospective nature of study, exemption was granted from Institutional Review Board of Shaukat Khanum Memorial Cancer Hospital and Research Centre.

Authors Contributions

Dr. Osama Shakeel (OS)-Study lead, Study concept and manuscript writing.

Dr. Aun Jamal Gill (AJG)-Data collection and analyses where needed.

Dr. Awais Naeem (AN)-Manuscript review.

Dr. Ihtisham Ul Haq (IUH)-Data collection and analyses where needed.

Dr. Faizan Ullah (FU)-Data collection and analyses where needed.

Dr. Masood Kant (MK)-Manuscript review.

Dr. Hannan Ali (HA)-Data collection and analyses where needed.

Dr. Warda Jabeen (WJ)-Manuscript writing.

Dr. Afzar Ali (AA)-Supervision of manuscript.

Dr. Khurram Mir (KM) -- Supervision of manuscript.

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