Primary melanoma of the female urethra: Case report of a very rare entity

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ABSTRACT
Melanoma is a malignant neoplasm that can affect any part of the body. The occurrence of melanoma in the female urethra is exceedingly a rare phenomenon with only one hundred and twenty-two (122) cases indexed since 1966. Here, a case of melanoma arising from the female urethra was reported. The case was associated with a 55 year old female admitted to our hospital with hematuria and swelling at her genital area. Physical examination showed 4 × 4.5 cm friable mass with irregular borders originating from the urethral meatus; otherwise there were no associated skin lesions. Biopsy of the lesion confirmed the diagnosis of melanoma. There was no evidence of metastasis on further work up.

Keywords: Female urethral tumors, melanoma, urethral neoplasms.

INTRODUCTION
Melanoma is a malignant neoplasm that can affect any part of the body. The occurrence in the female urethra is exceedingly a rare phenomenon with only one hundred and twenty-two (122) cases indexed since 1966 (Juan et al., 2011). The urethra is the most common site of origin in primary malignant melanoma of the genitourinary tract. Most malignant melanomas of the female urethra are located at the meatus or in the distal urethra.

Urethral melanoma shows its peak incidence in the older age group and the average patient’s age is 63 years. Patients with urethral malignant melanoma are admitted to the clinics with various complaints. Protruding mass is one of the most common clinically observed due to distal urethra more frequently involved than other parts of the body (Katz and Grabstald, 1976). The disease is three times more common in women than men (Ander et al., 1991; Reed, 1896). Reed (1896) reported the first case of primary malignant melanoma of the female urethra in 1896. The tumor is usually pigmented and varying in color from black to blue or light brownish lesions, which are firm, nodular and often ulcerated.

Grossly, the tumor may be easily confused with a caruncle (Nakamoto et al., 2007). Primary malignant melanoma of the female urethra tends to metastasize at the early stage through the superficial lymphatics to the vulva and vagina by the deep lymphatics to the inguinal lymph nodes and occasionally to distant sites by the haematogenous route.

Survival of malignant melanoma depends on the stage, location and size of the neoplasm at the time of diagnosis. Histological characteristics do not affect the prognosis significantly and all histological types are treated in a similar manner (Iversen and Robins, 1980; Poore and Cullough, 1996). Despite major surgery, radiotherapy or immunotherapy, malignant melanoma usually has a poor prognosis (Nissenkorn and Marshak, 1987).

CASE REPORT
A 55-year-old Ethiopian woman was admitted to the urology ward due to a protruding mass in her genital area and hematuria. On physical examination, a 4 × 4.5 cm friable mass with irregular borders originating from the posterior wall of urethral meatus was observed (Figures 1 to 3). There were no inguinal lymphadenopathies.

The clinical examination revealed no sign of previous cutaneous biopsy or existence of recent suspicious pigmented lesion of other localization. Metastatic evaluation including computerized tomography scan of the chest, abdomen and pelvis as well as, chest X-ray revealed
Academic Web Journal of Medical Science; Tegene et al; 012

Figure 1: Distal urethral mass measuring 4 × 4.5 cm and friable with irregular borders found to be primary melanoma on pathologic exam.

Figure 2: Intraoperative picture of the vulvovaginal area demonstrating a 4 × 4.5 cm friable mass with irregular borders originating from the posterior wall of urethral meatus. Histopathologic study proved it to be primary malignant melanoma of the distal urethra.

no evidence of disseminated disease. Histopathologic analysis revealed a polypoid ulcerated tumor partially covered with squamous and transitional epithelia. The tumor was composed of loosely cohesive nests of a typical epithelioid and spindle shaped melanocytes showing diffuse and nested growth pattern. The neoplastic cells had abundant eosinophilic cytoplasm, large hyperchromatic nuclei with prominent nucleoli and high mitotic activity (15/10 HPF). Most of the tumor cells contained coarsely granular melanocytic pigment. Lymphovascular invasion was histologically seen. A diagnosis of primary malignant melanoma of the distal urethra was made and the tumor was removed by a local excision (Figures 4 to 6).

**DISCUSSION**

Melanoma is a malignant neoplasm that can affect any part of the body. The occurrence in the female urethra is exceedingly a rare phenomenon with only one hundred and twenty-two (122) cases indexed since 1966 (Juan et al., 2011). The urethra is the most common site of origin in
primary malignant melanoma of the genitourinary tract. Most malignant melanomas of the female urethra are located at the meatus or in the distal urethra (Katz and Grabstald, 1976).

Patients with urethral malignant melanoma are admitted to the clinics with various complaints. Protruding mass is one of the most common clinically observed due to distal urethra more frequently involved than other parts (Abdulkadir et al., 2011). Other symptoms may include urethral mass, non-specific perineal pain, dysuria, frequency, incontinence, hematuria or local bleeding.

Metastatic melanoma should be excluded and as such detailed history, careful examination of the skin and evaluation of other visceral primary sites are required to confirm the primary nature of the lesion.

Histological appearance includes the whole range seen at other primary sites, including diffuse, nested, fascicular

**Figure 3:** Distal female urethral mass measuring 4 × 4.5 cm friable, with irregular borders originating from the posterior wall of urethral meatus. The mass was found to be primary malignant melanoma on histopathologic studies.

**Figure 4:** Distal urethral mass measuring being excised along with part of posterior urethra.
and storiform patterns. Amelanotic melanomas cause the greatest diagnostic challenge, so epitheliotropism as well as, pagetoid spread suggested a primary melanoma.

Urethral melanoma should be differentiated from benign pigmented lesions including genital lentiginosis, atypical melanocytic nevi and atypical lentiginous hyperplasia. Differential diagnosis includes sarcoma, spindle cell carcinoma, small cell carcinoma, lymphoma and plasmocytoma (Cvjetko et al., 2010). Conventional histopathological prognostic factors (tumor depth, level of
invasion, mitotic rate and ulceration) do not play important role in predicting outcome and biological behavior in the primary malignant melanoma of the female urethra as in the primary skin melanomas (Abdulkadir et al., 2011).

Surgical intervention plays the major role in first line therapy in the management of melanoma of the urethra. Patients who have lesion size more than 1 cm ulceration of their lesion and lymphadenopathy should also undergo extended inguinal lymphadenectomy (Abdulkadir et al., 2011). Local relapses and systemic metastases frequently develop in the early post-operative period following the removal of primary urethral malignant melanoma. Therefore, surgery alone is not adequate to control local relapses and systemic metastases, thus, adequate post-operative adjuvant therapy is required to prevent the relapse and progression of the disease. Combined use of multiple chemotherapeutic agents such as cisplatin, dacarbazine, carmustine and tamoxifen are recommended in lieu of monotherapy.

Our patients had adverse histological features (high mitotic activity, ulceration and Lymphovascular invasion) and big sized tumor. Therefore, excision of the mass along with distal urethra was done without inguinal lymphadenectomy. Post-operative adjuvant chemotherapy was offered with no evidence of recurrence after 8 months post-operatively.

REFERENCES


