

Primary Malignant Melanoma of the Male Urethra

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Primary malignant melanoma occurring within the male urethra is very rare. Here we report a case of malignant melanoma of the urethra in a 74-year-old man. He presented with asymptomatic gross hematuria for 5 months. His glans penis and adjacent penile skin had become discolored black 10 years ago. Cystourethroscopy showed a smooth oval-shaped elevated mass in the fossa navicularis. There were no abnormal findings in the proximal urethra and urinary bladder. Computed tomography did not detect any inguinal lymph node enlargement or distant metastases. The patient underwent partial penectomy and ilioinguinal lymph node dissection. Grossly, the distal urethra revealed an ovoid pigmented nodule, that measured 1 × 0.5 cm. Microscopic findings showed a nodular malignant melanoma arising in the urethral mucosa with pagetoid spread to the epidermis of the glans penis. There were no recurrences over a period of 12 months after surgery without chemotherapy. This is the second case of a primary malignant melanoma of the male urethra in Korea.

Key Words : Urethra; Male; Melanoma

Primary malignant melanoma of the male urethra is rare and associated with a poor prognosis. The urethra and the penis are the most frequently involved sites of malignant melanoma in the male genitourinary tract. The outcome is dependent on early diagnosis and surgical intervention. Since the clinical presentation of urethral melanoma is similar to common urothelial carcinomas, for most cases there is a delay in diagnosis. A knowledge of various presentations associated with primary urethral melanoma, as well as its histopathological features, is an important component to early diagnosis and effective treatment.¹ Here we report a case of primary malignant melanoma of the male urethra with a brief review of the literature.

CASE REPORT

A 74-year-old man was admitted to our hospital because of gross hematuria for 5 months. The patient had a 10-year history of irregular black discoloration of the glans penis. Physical examination revealed a geographic pigmentation of the glans penis and penile skin with a small protruding hemorrhagic nodule in the urethral meatus (Fig. 1A). Physical examination

revealed no palpable mass in either inguinal area. An excisional biopsy of the mass was performed, which revealed infiltrating large tumor cells having brown melanin pigments. Cystourethroscopy revealed a reddish black-colored, non-papillary, oval-shaped tumor in the distal urethra, just lateral to the fossa navicularis (Fig. 1B). Computed tomography revealed no lymph node enlargement or distant metastasis. We made the diagnosis of primary malignant melanoma of the urethra. The patient underwent partial penectomy and ilioinguinal lymph node dissection. Serial sections of the distal urethra disclosed a slightly elevated grayish white ulcerating tumor, measuring 1 × 0.5 cm. The mucosal surface of the tumor showed black pigmentation. Microscopic findings showed nodular sheets of spindle or epithelioid cells, infiltrating into the stroma up to 5 mm (pT4) (Fig. 2A). The superficial tumor cells contained melanin pigments, but deep tumor cells were amelanotic (Fig. 2B). Fontana-masson staining revealed intracellular melanin. Immunohistochemical stains for human melanoma black (HMB)-45 (Fig. 2C) and S-100 protein revealed strong positivity of the tumor cells. The tumor extended to the urethral meatus and showed pagetoid spread to the penile skin (Fig. 3). Melanin pigment-laden macrophages were scattered in the dermis. The patient refused chemo-

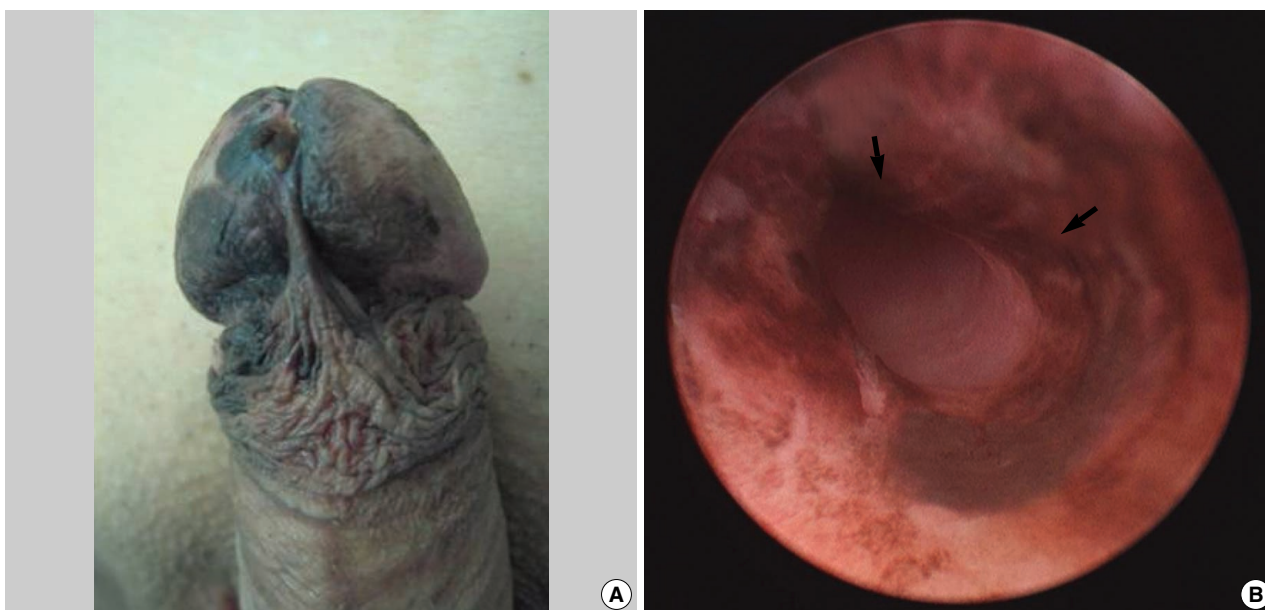


Fig. 1. (A) The patient shows geographic dark black pigmentation in his glans penis and penile skin. (B) Cystourethroscopic finding shows irregular dark black pigmentation and an elevated nodule in the urethral mucosa (arrows).

therapy. The patient was followed up for 12 months after surgery with no evidence of recurrence or lymph node metastasis.

DISCUSSION

Primary malignant melanoma of the genitourinary tract accounts for fewer than 1% of all malignant melanomas.² Melanocytes are of neuroectodermal origin, which is the reason for the low incidence of melanoma in endodermally derived urethral epithelium.

Primary melanoma accounts for approximately 4% of urethral tumors. Since Tyrell³ reported the first case of male urethral malignant melanoma in 1871, several case reports and small series have been described in the English language literature.¹⁻¹⁰ In Korea, there has been only one case reported of primary urethral malignant melanoma.¹¹ That case was a 77-year-old man who presented with turbid urine. He had a malignant melanoma measuring 1 cm in the proximal bulbous urethra without lymph node or distant metastasis. He was managed by transurethral resection and showed no recurrence at 6 months of follow-up.

Malignant melanoma of the urethra is three times more common in women than men. The distal urethra is affected most commonly in both sexes.³ In men, 55% occur at the fossa navicularis, 5% at the urethral meatus, 15% in the pendulous

urethra, 10% in the bulbous urethra and 15% in the prostatic urethra.⁷ The glans penis may also be involved in some patients.

The clinical presentation of urethral melanoma is similar to those of other urethral tumors, hematuria is the most common symptom. Some patients present with melanuria¹¹ or dark pigmented lesion, which is helpful for this diagnosis. There is often a considerable delay from the onset of symptoms to the patient being admitted to the hospital, the mean delay was 2 years in one review.⁷ The tumors are frequently polypoid and may be confused with urethral polyps, urethral caruncle, or more common malignant urethral tumors. They can also present as discrete or irregular macules along the urethral mucosa, or rarely, as an ulcerated lesion.²

On microscopic examination, urethral malignant melanoma shows a wide histologic spectrum, including diffuse, nested, storiform, and even pseudoglandular growth of the pleomorphic tumor cells.¹² The tumor cells are epithelioid or spindle shaped with or without melanin pigment. Amelanotic melanomas may cause particular diagnostic difficulties. The differential diagnosis of melanoma with sarcoma, spindle cell carcinoma, poorly differentiated urothelial carcinoma and extramammary Paget's disease is based on a careful search for melanoma *in situ* or melanin pigment and immunohistochemical stains including S-100 protein and HMB-45, and Melan A. Electron microscopy may also be helpful by demonstrating melanosomes in different

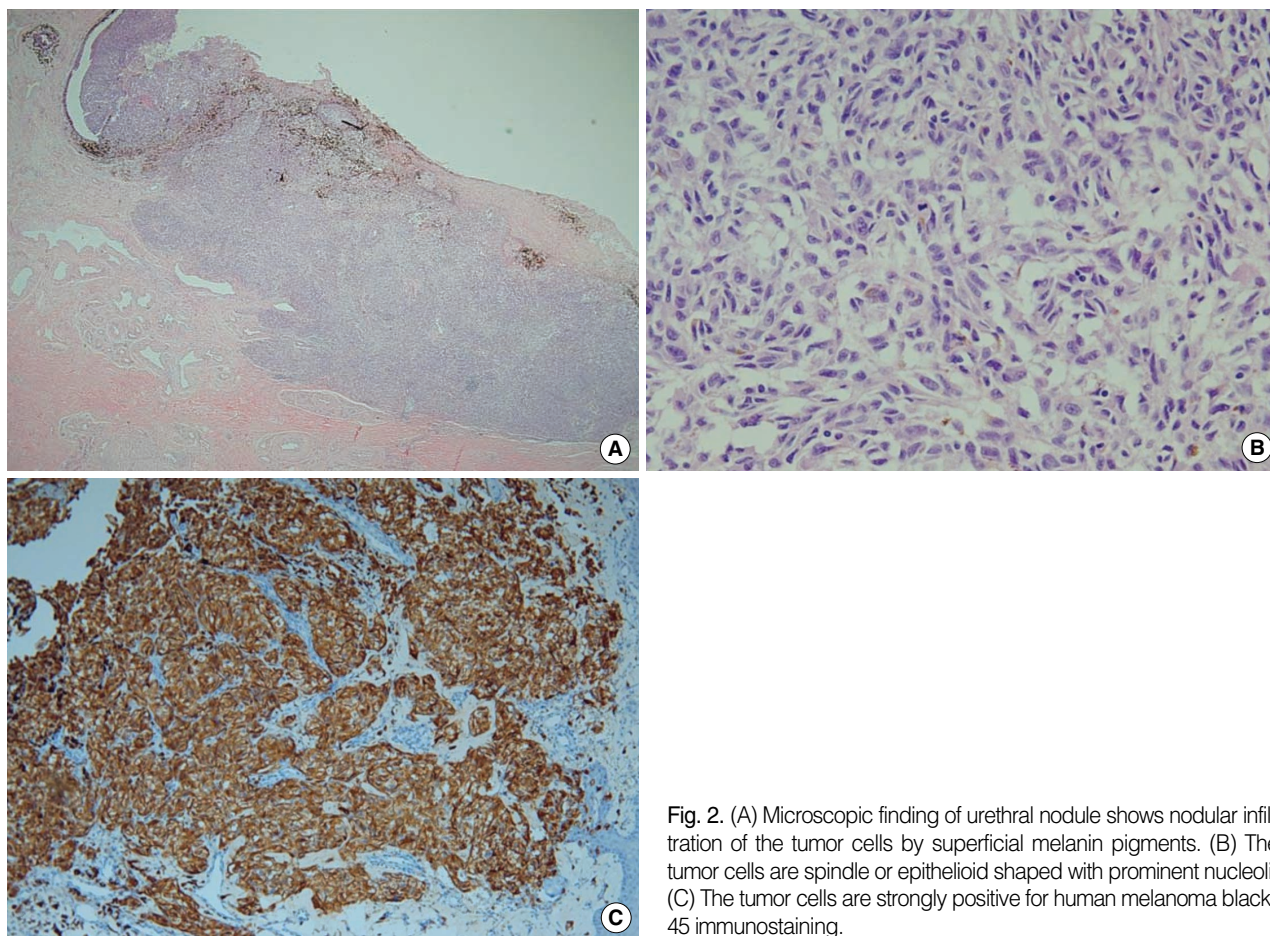


Fig. 2. (A) Microscopic finding of urethral nodule shows nodular infiltration of the tumor cells by superficial melanin pigments. (B) The tumor cells are spindle or epithelioid shaped with prominent nucleoli. (C) The tumor cells are strongly positive for human melanoma black-45 immunostaining.

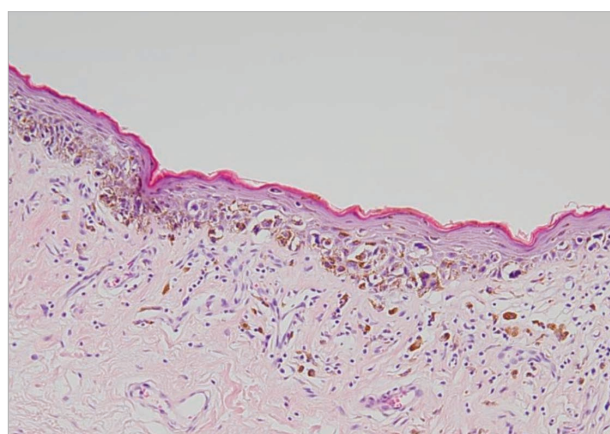


Fig. 3. The penile skin shows pagetoid spread of the tumor cells and melanophages in the dermis.

stages of maturation, especially in an amelanotic melanoma.

Urethral malignant melanoma has a worse prognosis than its cutaneous counterparts. At the time of diagnosis, most urethral melanomas are deeply invasive and local extension to the vagina, the corpora cavernosa or urinary bladder is common.¹³ Inguinal lymph node metastases or distant metastases are also present

from an early stage.² Therefore, partial or total penectomy with bilateral inguinal lymph node dissection has been recommended.⁷ The important poor prognostic factors in mucosal melanoma are tumor depth more than 3.5 mm, presence of ulceration and tumor diameter more than 15 mm.⁵ Robutti *et al.*¹⁴ reported that tumors invading to a depth less than 2 mm had an excellent prognosis. But the studies of Oliva *et al.*² showed that conventional prognostic factors, such as depth of invasion or tumor stage, did not have as notable a role in predicting survival in mucosal melanoma. In our patient, the tumor invaded to a depth of 5 mm and there was no evidence of lymph node or distant metastasis. The patient underwent partial penectomy with lymph node dissection. He was free of disease at 12 month.

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