



Carcinoma in situ of the external urethral meatus: Case report and literature review

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Abstract

Introduction: Primary tumor of the urethra in male is a rare case, occurred <1% of urologic malignancies and occurs more often in female.

Material and Methods: We reported a case of carcinoma insitu of the external urethral meatus on 27 years old male who had mass excision and fulguration.

Results: Patient 27 years old male came with chief complaint painless bloody urine, that came out at the beginning of urinate since 1 month before admission. There were no history of passing stone or palpable lump at penile. On physical examination we found a mass at size of 5 mm and 3 mm at the external urethral meatus, it consistency was soft, raw surfaces and painless. There were no inguinal lymphadenopathy, and digital rectal examination within normal limit. On urinalysis examination we found a lot of erythrocytes, while other laboratories finding were within normal limits. Patients had been performed diagnostic urethrocystoscopy, we found a mass at external urethral meatus, while the urethra, prostate, bladder neck, bladder and both ureter's orifice were no abnormality. we performed mass excision and fulguration and the results of histopathologic examination (P.16.09.2756) dated on 24th September 2016 was carcinoma in situ of the external urethral meatus. At post-operative day 30, patient came without any complaints, spontaneous urination was clear yellowish and on physical examination there was no lesions at the external urethral meatus.

Conclusion: Primary urethral tumor in male patients is a very rare case, compromising only less than 1% of total malignancy cases in urology and usually more common in female patients. For lesion which involve more proximal part of anterior urethral, excision of urethral segment, with corpus penis preservation, might be decent enough for superficial tumor.

Keywords: carcinoma in situ, external urethral meatus, initial gross hematuria

Introduction

Primary urethral tumor in man is a rare case, comprising less than 1% of total incidence of urology malignancies dan usually more common in women. Primary urethral tumor is a tumor which first time detected in the urethra, different from secondary tumor which is found after recurrence which previously has been diagnosed and undergone tumor treatment in the other urinary tract. Distal urethral tumor tends to show a lower stadium stage and has a better prognosis, which is an important prognosis indicator. Recently, penis preservative surgery was performed in a part of distal urethral cancer in male patients. The main symptoms are urethral bleeding/initial gross hematuria, palpable urethral mass and obstruction when urinating [1, 2, 3, 4, 5].

Primary urethral cancer is considered as a rare cancer, comprising less than 1% of total incidence of malignancies. In early 2008, the prevalence of urethral cancer in 27 countries as part of European Union was 4,292 cases with the number of incidences are 655 new cases annually. Tumor in man urethra is grouped based on the location and histopathology. Around 60% urethral tumor can be found in bulbomembranous, 30% in penile urethra, and 10% in urethra prostatic. Tumor histopathology of urethra pars bulbomembranous is consisted of 80% squamous cell, 10% transitional cell and 10% adenocarcinoma or undifferentiated.

Tumor histopathology of penile urethra (anterior) is consisted of 90% squamous cell, and 10% transitional cell, while in urethra pars prostatic is consisted of 90% transitional cell and 10% squamous cell [1, 2, 3].

Etiology of urethral cancer is still not clear. Although smoking, amine aromatic exposure, and analgesic abuse associated with bladder transitional carcinoma cell, there was no direct correlation between those factors with urethral carcinoma appearance. However, patients with bladder cancer history can increase the risk of urethral cancer occurrence. Other etiologies are including chronic inflammation caused by sexual transmitted disease, urethritis, and urethral stricture, and apparently these factors also play a role in causing human papilloma virus (HPV) as the cause of urethral squamous cell carcinoma. Several studies has suggested that infection and chronic irritation as potential etiologies in tumorigenesis of this urethral malignancy. Kaplan et al (1967) found that 37% man with urethral cancer has several histories of venereal disease. Moreover, Ray et al found a 44% association between urethral cancer and the history of sexual transmitted disease. Human papillomavirus (HPV) also associated with several cases of urethral cancer. Wiener et al (1992) showed that the availability of DNA HPV in 4 patients

(29%) was found in 14 cases of primary urethral cancer [1, 3, 4, 13]. Urethral carcinoma in male patients can extent with a direct expansion between the adjacent structures, usually involving vascular cavities from corpus spongiosum and periurethral tissue, or can be metastasized by performing embolization lymphatic to the regional lymphoid. Drainage will be performed by the lymphatic system from anterior urethral to lymphoid inguinal superficial and deep, usually to lymphoid iliac externa. Tumor from urethra posterior usually will be expanded to the lymphoid pelvic. Lymphatic inguinal cancer enlargement can be palpable in 20% cases and almost always related with metastatic tumor, contrary with urethral penile tumor, while the palpable lymphatic cancer may be caused by inflammation process. Hematogenous expansion rarely happen, except in advance stage [1, 3].

The initial symptoms may be including hematuria or bleeding urethral discharge, comprising 62% of total cases. Other symptoms in more advance stages are including mass outside the urethra (52%), outlet obstruction of the bladder (48%), pelvic pain (33%), fistula urethrocutan (10%), and abscess formation (5%). The most common symptoms are urethral bleeding/initial gross hematuria, palpable urethral mass and obstruction sign while urinating [1, 6].

In male patients, physical examination should perform starting from inspection, lesion existence in meatus urethral externus and also lump or mass existence in the penis. The examination then continued by palpating in genitalia externa for induration or mass suspicion. Palpation in inguinal area is performed bilaterally to assess lymphatic cancer enlargement, location, size and mobility. Then digital rectal is performed to assess the extension to anorectal area [7]. The role of urine cytology assessment in urethral primer tumor is limited, and the sensitivity is around 55-59%. Detection results are dependent to the histopathology, in male patients, the sensitivity of urothelial carcinoma is 80%, while for squamous cell carcinoma is only 50% of total cases [8, 15].

Urethrocystoscopy and biopsy for diagnostic are performed to assess urethral primer tumor in how far the tumor has developed, location and the underlying histopathology. To accurately assess the histopathology, healthcare professionals will need to do adequate surgery procedure, incite biopsy (urethral proximal or distal) should be marked and posted together with tumor clinical information to pathologist [1, 15].

The main treatment of primary urethral tumor is surgery. In general for anterior urethral carcinoma, it is more recommended to perform surgery, with a higher prognosis than posterior urethral carcinoma, which usually more relate to local expansion and distant metastatic. Transurethral resection, local excision, or distal urethrectomy and perineal urethrectomy are accepted as treatment options for patients with superficial tumor, papillary or low grade. In rare case, malignancy which only associated with urethral mucosa (stadium 0/Tis, Ta), resection and ulguration can be used as treatment options. Penis amputation is used for a deeper lesion infiltration. Traditionally, 2 cm of tumor proximal limit is used, although there are no study performed to determine the most optimum margin. The number of recurrences is quite low in pre-invasive lesion; however the failure rate is higher in more invasive tumor. Currently, laser is the best treatment option for superficial lesion [1, 3, 9, 10, 11, 12, 14, 15]. The work has been reported in line with the SCARE criteria [16].

Objectives

We reported a case of external urethral meatus carcinoma in a male patient, 27 years old which already underwent excision of tumor mass and fulguration, where on diagnostic urethroscopy did not found any abnormality in anterior and posterior urethral and bladder. Primary urethral tumor in external urethral meatus is a very rare case and usually happen in female patients, in our study, this is the first case in our institution. In the more common case, urethral tumor has already reach an advance stage and infiltrated.

Case report

A male patient, aged 27 years old, came to our institution complaining bleeding during the beginning of urination (initial gross hematuria) since 1 month before admission. Complaint was not followed with pain during urination. The patient refuse to experience of passing stone and cloudy urine, patient also did not complain about a mass in urinary tract. There was history of urinary tract infection. The history of a frequent sex partner change and suffering from sexual transmitted disease were denied. He also denied the previous history of penile mass.

In the physical examination, we found 5 mm and 3 mm masses near external urethral meatus, mild in consistency, raw surface and unpainful. He did not experience inguinal lymphatic enlargement, and digital rectal examination within normal limit. In urine analysis, we found a high number of erythrocyte and leucocyte, while the other laboratory test showed normal values. Patient underwent diagnostic urethroscopy. During urethroscopy, we found a mass in external urethral meatus, while alongside of the urethral, prostate, bladder neck, bladder and both ureteral opening did not found any abnormalities. We decided to perform mass excision and fulguration, with histopathology result (P.16.09.2756) in 24th September 2016 showed in situ carcinoma of external urethral meatus.

Patients underwent surgery with general anesthesia. Blood pressure before surgery was 120/70 mmHg, heart rate before surgery was 84 times/minute, normal electrocardiogram with SpO2 100%. Laboratory preoperative value of hemoglobin was 12.4 g/dL, sodium 135 mEq/L, potassium 4.8 mEq/L, creatinine 0.60 mg/dL, urea 30 mg/dL, random blood glucose was 102 mg/dL. Irrigation liquid used was sterile Aqua, 80 cm above the surgery table. In lithotomy position, diagnostic urethroscopy was perforfed, we found masses in external urethral meatus, anterior and posterior urethral, bladder neck, bladder, and both ureteral opening within normal limit. We decided to perform mass excision and fulguration. On 30th post-operative day control, patient came without any complaint, spontaneous urination was in clear yellowish and in physical examination, we found no lesion in the external urethral meatus.



Fig 1: Mass in external urethral meatus

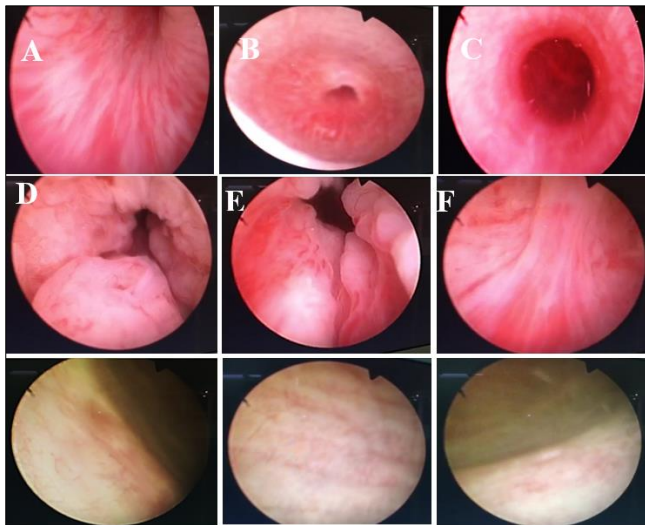


Fig 2: Urethroscopy. A. urethra pars pendulare, B. urethra pars bulbosa, C. urethra pars bulbomembranosa, D. verumontanum, E. prostate (prostatitis impression), F. bladder neck, G. right ureteral opening. H. left ureteral opening, I. posterior wall of bladder.

Type of examination	: Biopsy
Number	: P.16.09.2756
Macroscopic:	four pieces of tissue the largest diameter is 0.5 cm, the smallest diameter is 0.3 cm, the color is brownish white
Macroscopic:	External urethral orifice are partially coated by hyperplastic flaky layered epithelium. large cell nucleus, polymorph, hyperchromatic, mitosis found basalis membrane is still good, partially coated by transitional epithelium, the nucleus is normal. subepithelial seen with fibrocollagen connective tissue with lymphocytes, no malignant tumor cells found.
Conclusion:	carcinoma in situ external orifice of urethral

Fig 3: Histopathology result.

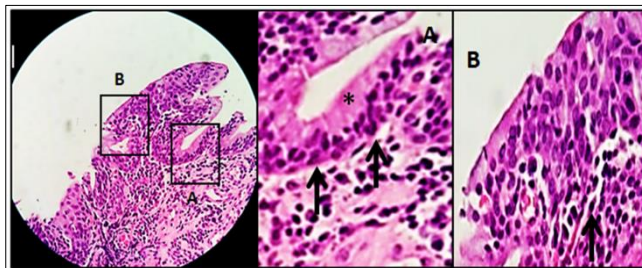


Fig 4: Histopathology picture. A. showing basalis membrane which is still looks good (small nucleus, regular edge, lot of cytoplasm). B. showing abnormality epithelial thickness (enlargement of nucleus, few cytoplasm). Mark↑basalis membrane, (*): cytoplasm.



Fig 5: External urethral meatus after 3 months follow-up.

Discussion

Etiology factor of urethral carcinoma including chronic

inflammation caused by sexually transmitted disease which usually, urethritis, and urethral stricture, and apparently these factors also play a role to allow human papilloma virus as the cause of squamous cell carcinoma from urethra [1, 3, 4, 13]. In this patients, the possible cause might be from urethritis, since the anamnesis showed there was history of urinary tract infection, but the history of a frequent sex partner change and suffering from sexual transmitted disease were denied. He also denied the previous history of penile mass.

The most common symptoms are urethral bleeding, initial gross hematuria, palpable urethral mass and obstruction symptoms during urinating.^{1,6} In this patient, the main symptom was bleeding during the first time of urinating (initial gross hematuria) which is not followed with pain since 1 month before hospital admission.

Urethroscopy and biopsy diagnostic are allowing primary urethral tumor assessment to determine how far tumor has expanded, location and the underlying histopathology.^{1,15} Commonly for urethral carcinoma anterior, the recommended treatment option is surgery, with a better prognosis than posterior urethral carcinoma [1, 2, 3, 4, 5, 9, 10, 11, 12, 14]. In this patient, we performed diagnostic urethroscopy, when we found a mass in meatus urethral externus, while in alongside the rest urethral part, bladder and both ureteral opening, we found no abnormalities. We had performed mass excision and fulguration and histopathology assessment was in situ carcinoma in the opening of external urethra.

Conclusion

Primary urethral tumor in male patients is a very rare case, compromising only less than 1% of total malignancy cases in urology and usually more common in female patients. Anterior urethral tumor tends to show a milder stage and has a better prognosis. Anterior urethral carcinoma should be treated by surgery, with a better prognosis than posterior urethral carcinoma. In the rare case which only associated with urethral mucosa (stadium 0/Tis, Ta), resection and fulguration can be used as treatment options. For lesion which involve more proximal part of anterior urethral, excision of urethral segment, with corpus penis preservation, might be decent enough for superficial tumor.

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