Case Report

Primary male urethral squamous cell carcinoma presenting with a genital abscess

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Abbreviations & Acronyms

- BSC = best supportive care
- Cx = chemotherapy
- nd = not described
- Rx = radiation therapy

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Received 22 January 2019; accepted 19 May 2019. Online publication 13 June 2019 **Introduction:** Primary urethral cancer is a rare disease accounting for <1% of all urological cancer cases. We encountered a patient with urethral squamous cell carcinoma diagnosed during treatment of a genital abscess.

Case presentation: A 69-year-old male was referred to our hospital for recurrent urethritis and swelling of the scrotum. The genital skin was atrophic with a purulent discharge. A diagnosis of epididymitis-induced genital abscess was made, and conservative treatment was administered. However, local infection recurred and the infected tissue was resected 4 months after the first examination. The pathological diagnosis was squamous cell carcinoma. Subsequently, radical surgery was performed, but the patient died 17 months postoperatively.

Conclusion: Four patients with primary urethral squamous cell carcinoma presenting as genital abscess have been reported. Careful examination is necessary while considering a malignant tumor as an underlying disease.

Key words: perineal abscess, recurrent urethritis, scrotal abscess, squamous cell carcinoma, urethral cancer.

Keynote message

We experienced a 69-year-old male with primary urethral squamous cell carcinoma presenting with a genital abscess. A genital abscess may occur as an initial symptom of urethral cancer. Careful examination is necessary while considering a malignant tumor as an underlying disease.

Introduction

Primary urethral cancer is a rare disease accounting for <1% of all urological cancer cases.¹ We encountered a patient with urethral squamous cell carcinoma diagnosed during treatment of a genital abscess. We investigated the case and previously reported similar cases.

Case presentation

A 69-year-old man with pain on urination visited a physician. The patient had no contributory illnesses except hypertension. Urethritis was diagnosed and treated with an antibiotic agent, but it recurred and swelling of the scrotum over the perineum developed. The patient was referred to our department and admitted for further examination and treatment. His general condition was fair, and no high fever developed. Swelling of the scrotum over the perineal region was noted; the perineal skin was disintegrated and there was a purulent discharge. The scrotal skin was partially necrotized, but no tumorous changes were seen. The prostatic findings were normal.

Blood and chemistry test results on admission were within normal ranges, except for a mild elevation in C-reactive protein. A urinary sediment examination showed a mild degree of pyuria, but no bacteria were observed. Enhanced computed tomography found ulceration



Fig. 1 Enhanced computed tomography axial and sagittal slices showing fluid collection accompanied by gas (arrows) in the scrotum over the perineal region at the first examination. The mass in the perineal region enlarged thereafter. (a) First examination. (b) Two months after first examination. (c) Five months after first examination.

| Fig. 2 Macroscopic and microscopic findings of |
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| primary urethral cancer. (a) Macroscopic finding |
| at 4 months after first examination. The infected |
| tissue in the perineal region was resected, and a |
| diagnosis of squamous cell carcinoma was made. |
| (b) Macroscopic finding at 6 months after first |
| examination. (c) Microscopic finding of urethral |
| squamous cell carcinoma with slight cornification |
| (hematoxylin and eosin staining). Arrows show |
| the cornification area. |

| | Age | Local finding | Duration [†] | Outcome | Treatment | Author |
|---|-----|----------------------|-----------------------|---------------|-----------------|------------------------|
| 1 | 68 | Periurethral abscess | nd | nd | Removal, Rx | Dickinson⁵ |
| 2 | 48 | Periurethral abscess | 6 months | 3 month dead | BSC | Angulo ⁶ |
| 3 | 63 | Fournier's gangrene | 7 months | 6 month dead | Removal | Matsumura ⁷ |
| 4 | 40s | Fournier's gangrene | 18 months | 24 month dead | Removal, Cx, Rx | Moore ⁸ |
| 5 | 69 | Scrotal abscess | 5 months | 17 month dead | Removal, Cx, Rx | Ours |

†Until diagnosis from the first presentation.

accompanied by gas in the scrotum over the perineal region. The cavernous portion was lightly stained in the early phase, and a region with a lower concentration than the surrounding area was noted in the late phase (Fig. 1a). The patient was diagnosed with an epididymitis-induced scrotal and perineal abscess. A skin incision was made in the abscess region; and discharge of pus, wound irrigation, and Cx with ampicillin sodium were performed. The local findings improved and the perineal wound was sutured. After 3 weeks, the patient re-

visited due to urinary incontinence from the perineal suture wound. Endoscopic examination showed rude tissues protruded into the urethral lumen, and causing urethral stricture. Posterior urethral stricture and urethrocutaneous fistula were diagnosed and percutaneous cystostomy was performed. Urine cytology was negative. One week later, a purulent discharge from the suture wound and external urethral meatus was observed. Re-infection was diagnosed and wound opening, pus drainage, and Cx with levofloxacin were performed.



Fig. 3 The clinical and therapeutic course. Although postoperative Rx was performed, distant metastases developed. The response to Cx was short, and the BSC policy was selected.

On enhanced computed tomography 2 months after the first examination (Fig. 1b), discharge of pus from a deep region was insufficient and resection of morbid granulation was considered necessary (Fig. 2a). The infected tissue in the perineal region was resected under general anesthesia. Morbid granulation and scarring reached from the cavernous portion to the urethral mucosal surface. It was not possible to entirely resect the infected tissue. On pathological examination, proliferation of cornified atypical cells was observed and he was diagnosed as squamous cell carcinoma. No finding of metastasis was noted on careful examination, including enhanced computed tomography. The tumor in the perineal region rapidly grew thereafter (Figs 1c,2b). Urethral tumor resection and pelvic lymph node dissection were performed based on the diagnosis of primary urethral cancer. Being continuous to the mass in the perineal region, the penis was entirely hardened excluding the glans. The tumor was en bloc excised including the entire urethra. The pubis was strongly adhered, suggesting invasion. The bilateral external iliac lymph nodes and obturator lymph node were dissected. Tumor size in the cross-section of the excised specimen was 11×5.5 cm. On microscopy, atypical squamous cells with a cornification tendency were present, and the tumor was diagnosed as squamous cell carcinoma (Fig. 2c). The stump was positive, and all lymph nodes were negative. Since the resected stump was positive, 54 Gy irradiation was applied to the pelvic and perineal regions. However, lung metastasis and local recurrence occurred. Cx (3 weeks schedule; paclitaxel 175 mg/m² Day 1, ifosfamide 1200 mg/m² Days 1-3, cisplatin 25 mg/m² Days 1-3) was administered, but his general condition gradually worsened, and he died 17 months after radical surgery.

Discussion

Primary urethral cancer is a rare disease and is reported to account for <1% of all urologic cancer cases.¹ The incidence is higher in males and onset age is commonly in the 60s in both males and females. Regarding the histologic type, urothelial cancer is most frequent, accounting for 47–65% of cases, while squamous cell carcinoma accounts for 16–30%,^{2–4} although squamous cell carcinoma was the most frequent in males and females in one study.¹

In the present case, a resected sample could not help identify the origin of the squamous cell carcinoma. However, we diagnosed primary urethral cancer because of an endoscopic finding in the urethral lumen and the skin appearance in the scrotum in the early stage, although a primary skin malignancy was among the differential diagnoses.

Four cases of male urethral squamous cell carcinoma diagnosed after abscess formation in the genital region were previously reported. Five cases, including our case, are shown in Table 1. All cases were accompanied by severe local infection, and in two cases Fournier's gangrene was present. The other cases were also treated with antimicrobial agents and debridement, but the disease was intractable. Four cases were complicated by an urethrocutaneous fistula, and cystostomy was performed to treat urethral stenosis or fistulation in all cases. Furthermore, the following common points were observed and should be considered: the urethral tumor was not diagnosed during treatment of a genital abscess in any case, and was only diagnosed after protracted infection and/or later local swelling. According to reports, several months passed before diagnosis in many cases. In our patient, 5 months were required to diagnose the urethral cancer (Fig. 3). Urine cytology was performed during treatment of the scrotum abscess, but a histological examination was necessary at the time of debridement. Since a genital abscess may develop with urethral cancer as the underlying disease, careful examination is necessary while also taking a malignant tumor into consideration. The sensitivity of urine cytology in detecting urethral squamous cell carcinoma is low compared with that for urothelial carcinoma, and was reported to be approximately 50% in males.² Histological examination may be useful when debridement or urethroscopy is performed.

Conclusion

A patient with primary urethral squamous cell carcinoma diagnosed during treatment of a genital abscess was reported. Four similar cases were reported and some time was taken to diagnose the cancer. Careful examination is necessary while considering a malignant tumor as an underlying disease.

Conflict of interest

The authors declare no conflict of interest.

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