

Squamous Cell Carcinoma in the Meatus of a Distal Hypospadias

Makoto Imura, Shoichi Sasaki, Yoshiyuki Kojima, Hideyuki Kamisawa, Yutaro Hayashi, Kenjiro Kohri

Department of Nephro-urology, Nagoya City University, Graduate School of Medical Science, Nagoya, Japan

Submitted September 9, 2009 - Accepted for Publication November 17, 2009

ABSTRACT

A 39-year-old male presented with a tumor in the urethral orifice. A papillary tumor (1 cm × 1 cm) was found at the meatus of a distal hypospadias. The patient underwent tumor resection without urethroplasty. The pathological diagnosis was squamous cell carcinoma. No recurrence or metastasis was found during 2 years of follow-up. Squamous cell carcinoma in the urethral orifice of hypospadias is extremely rare. To the authors' knowledge, this is only the second case reported in the literature.

KEYWORDS: Urethral cancer; Squamous cell carcinoma; Meatus; Hypospadias

CORRESPONDENCE: Yoshiyuki Kojima MD, Department of Nephro-urology, Nagoya City University Graduate School of Medical Sciences, 1 Kawasumi, Mizuho-cho, Mizuho-ku, Nagoya 467-8601, Japan (ykojima@med.nagoya-cu.ac.jp).

CITATION: *UroToday Int J.* 2009 Dec;2(6). doi:10.3834/uj.1944-5784.2009.12.13

INTRODUCTION

Primary urethral carcinoma is a very rare condition and information in the literature is relatively limited. The authors describe a 39-year-old male with squamous cell carcinoma (SCC) in the meatus of a distal hypospadias. *Hypospadias* is a congenital anomaly of defective closure of the urethra, in which the meatus is located abnormally along the ventral side of the penis. It is one of the most common malformations in males, with a reported incidence of 5 to 8 per 1000 live male births [1]. To the authors' knowledge, this is only the second report of SCC associated with hypospadias in the literature.

CASE REPORT

A 39-year-old male with a distal hypospadias was referred to the authors' department due to a tumor in the urethral orifice. The tumor had been noticed 1 year before the patient's hospital visit. A papillary tumor (1 cm × 1 cm) was detected in the meatus of the distal hypospadias (Figure 1).

The patient had not undergone surgical repair for the hypospadias. His serum SCC antigen was normal (0.5 ng/mL). No metastatic focus was found on chest, abdominal, or pelvic

computed tomography (CT) scans. Cystourethroscopy did not reveal any abnormality in the bladder or proximal urethra.

The patient underwent tumor resection with a 1 cm surgical margin under spinal anesthesia. Urethroplasty was not performed. Immediately after tumor resection, urethroscopy was repeated under the same anesthesia to confirm complete tumor resection.

Pathological examination revealed squamous cells with increased nuclear variation and scattered cancer pearl cells (Figure 2). The final pathological diagnosis was well-differentiated SCC with chronic inflammation. There was invasion of subepithelial connective tissue but not the corpus cavernosum or periurethral muscle. The resection margins were tumor-free.

The patient was followed for 24 months. No metastasis occurred and no recurrence was found through urethroscopy. Follow-up chest, abdominal, and pelvic CT scans at 24 months showed no metastatic focus. Serum SCC antigen (0.5 ng/mL) did not increase postoperatively. He was able to urinate comfortably and had normal sexual activity.

Figure 1. A Papillary Tumor (1 cm × 1 cm) Detected in the Urethral Opening of a Distal Hypospadias.

doi: 10.3834/uij.1944-5784.2009.12.13f1



DISCUSSION

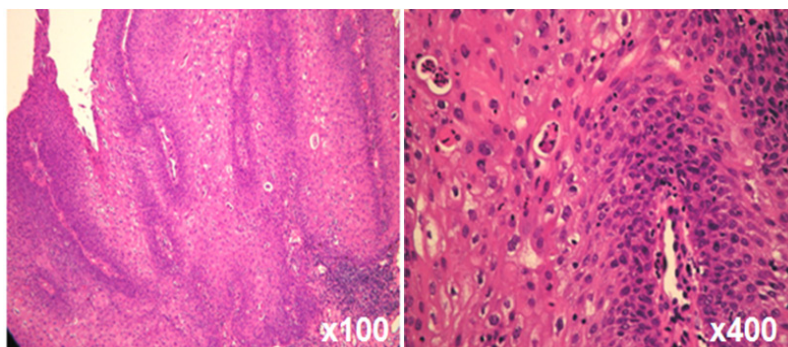
Carcinoma of the urethra is a rare urologic malignancy, accounting for less than 1% of all urologic malignancies. Although about 80% of male urethral cancers are squamous cell carcinoma, the histologic subtype of urethral cancer also varies by anatomic location. Carcinoma of the penile urethra is of squamous cell origin in 90% [2]. The SCC antigen is a useful marker to detect the recurrence and metastasis of penile SCC [3], although serum SCC antigen did not increase postoperatively in

the present case. Laniado et al [4] reported that an elevated SCC antigen level had a sensitivity of 57% and a specificity of 100% for nodal metastases in men with SCC of the penis.

Dodd et al [5] reported the first case of a 43-year-old male with SCC in the urethral orifice of penile-type hypospadias. Pomara et al [6] reported a case of a locally advanced verrucous scrotal cancer in a 68-year-old male with a penoscrotal hypospadias. The authors suggested that hypospadias and secondary chronic inflammation of the scrotal skin, caused by prolonged contact with urine, may contribute to an increased risk of verrucous carcinoma.

Kirkman [7] reported urethral carcinoma following urethroplasty. In the present case, the authors did not perform urethroplasty because the patient had no complaint of meatal dislocation and they wanted to avoid local recurrence in the neourethra after hypospadias repair. The identified etiologic factors of urethral cancer include chronic inflammation due to a history of sexually transmitted disease, urethritis, trauma, and urethral stricture. Complications of hypospadias repair, regardless of the repair method, include meatal stenosis, urethral stricture, wound infection, and impaired healing. Mild and localized infection can occur after hypospadias repair because of compromised vascularity, humidity, high temperature, and proximity to a potentially contaminated area. Even if no complication occurs, acute inflammation is always observed immediately after hypospadias repair. These adverse events may induce local recurrence in the neourethra, although there is no evidence to show this correlation. The authors believe that hypospadias should not be repaired in patients with urethral cancer, even if they wish to have it corrected.

Figure 2. Squamous Cells Showing Increased Nuclear Variation and Cancer-Scattered Pearl Cells (Hematoxylin and Eosin Stains). doi: 10.3834/uij.1944-5784.2009.12.13f2



REFERENCES

- [1] Belman AB. Hypospadias and other urethral abnormalities. In: Kelalis PP, King LR, Belman AB, eds. *Clinical Pediatric Urology*. 3rd ed. Philadelphia PA: WB Saunders; 1992:619-638.
- [2] Machele Donat S, Cozzi PJ, Herr HW. Surgery for penile and urethral carcinoma. In: Walsh PC, ed. *Campbell's Urology*. 8th ed. Philadelphia PA: WB Saunders; 2002:2983-2999.
- [3] Touloupidis S, Zisimopoulos A, Giannakopoulos S, Papatsoris AG, Kalaitzis C, Thanos A. Clinical usage of the squamous cell carcinoma antigen in patients with penile cancer. *Int J Urol*. 2007; 14:174-176.
- [4] Laniado ME, Lowdell C, Mitchell H, Christmas TJ. Squamous cell carcinoma antigen: a role in the early identification of nodal metastases in men with squamous cell carcinoma of the penis. *BJU Int*. 2003; 92:248-250.
- [5] Dodd M, Lawson P, Hayman J. Squamous cell carcinoma of the distal urethra in a patient with congenital hypospadias. *Pathology*. 1996; 28: 96-97.
- [6] Pomara G, Pomara S, Travaglini F, Maras L, Selli C. Verrucous scrotal carcinoma in a patient with hypospadias: is there a possible association? *Urology*. 2003; 61:224.
- [7] Kirkman NF. Urethral carcinoma following urethroplasty. *Br J Surg*. 48; 508-511, 1961.