Case report

Keratinizing desquamative squamous metaplasia in the kidney of a patient with complex spinal cord injury

Jack Coode-Bate, Melissa Clare Davies, Ian Cook

Salisbury District Hospital, NHS Foundation Trust, Salisbury, UK

Context: Keratinizing desquamative squamous metaplasia (KDSM) of the upper urinary tract was previously thought, but never proven, to represent a premalignant condition. However, no clear guidelines exist regarding the long-term management and follow up once this rare diagnosis has been made.

Findings: Patients with spinal cord injury often develop a range of risk factors for malignancy of the urinary tract. We present the first reported case of KDSM in the kidney of a 44-year-old man with spinal cord injury whose complex urological history includes previous augmentation cystoplasty, recurrent calculi, infections and indwelling catheters.

Clinical relevance: Recent literature has suggested a move away from the previous mainstay of treatment with radical surgery towards nephron-sparing endoscopic and radiological surveillance. This case highlights the complexity of preserving renal function in a spinal cord injured patient with KDSM of the kidney.

Keywords: Keratinizing desquamative squamous metaplasia, Kidney, Spinal cord injury

Case

A 44-year-old man suffered a complete (ASIA classification A) C4 spinal cord injury (SCI) over 22 years ago. For the first two decades his bladder was managed with a series of both urethral and then suprapubic indwelling catheters. Over this time he has produced recurrent renal calculi formed of 100% calcium phosphate, requiring multiple percutaneous nephrolithotomy and ureteroscopic interventions. In addition he has presented acutely on numerous occasions with significant urinary tract infections requiring intravenous antibiotics; microbiology samples have separately isolated *pseudomonas*, *enterococcus* and *proteus* in urine and/or blood cultures in the last year alone. Recently, all elective procedures to treat his stones have required careful monitoring and cardiorespiratory support on the intensive care unit for a minimum of 48 hours post-operatively.

Concurrent to his stone disease, the patient suffered progressively severe episodes of autonomic dysreflexia. As demonstrated on urodynamic studies, these were clearly associated with neurogenic detrusor overactivity of his bladder, which steadily became refractory to

management with oral anticholinergic agents and intravesical injection of botulinum toxin. Last year he underwent an elective augmentation cystoplasty with resiting of suprapubic catheter; a decision made according to patient preference over ileal loop urinary diversion and with the known requirement for ongoing endoscopic access and stone management.

A recent flexible ureteroscopy for calculi in the lower pole calyx of the left kidney (Figs. 1 and 2) revealed a moderate amount of blood clot and debris. The two stones were identified and fragmented with an holmium laser before extraction. In the upper pole calyx the abnormal appearance of keratin was noted and cold cup biopsies were taken from the underlying mucosa, which appeared macroscopically abnormal.

Histological and cytological analysis of the samples taken from the left kidney has confirmed the presence of keratin indicative of underlying squamous metaplasia (KDSM), but no evidence of dysplastic or malignant cells (Figs. 3 and 4).

Retrospective review of contrast-enhanced CT imaging demonstrated the evidence of cortical thinning and bilateral renal scarring associated with recurrent infections. However, no soft tissue mass or filling defects are visible in either kidney.

Correspondence to: Jack Coode-Bate, Urology Department, Salisbury District Hospital, Odstock Road, Salisbury, Wiltshire, SP2 8BJ, UK. Email address: jcoodebate@doctors.org.uk.

Figure 1 Coronal CT image demonstrating the calculus in the left left lower pole calyx and thinning of the cortex of the left kidney. The right kidney demonstrates some residual contrast and the evidence of a ureteric stent following previous right-sided ureteroscopy.

Discussion

KDSM of the upper urinary tract remains a rare histological finding. The first case report of this condition was published in 1882.¹ Although no clear etiology exists, associations have previously been made with chronic inflammation due to recurrent or persistent infections, stone disease and irritants such as cigarette smoke.²

Keratinizing squamous metaplasia in the bladder is thought to represent a pre-malignant condition that requires regular cystoscopic surveillance.³ Features associated with an increased risk for malignancy include the presence of dysplastic cells and larger volume keratin production. In KDSM of the renal

Figure 2 Axial CT image of the abdomen at the level of the lower pole left renal calculus.

Figure 3 Images from histopathology slides of cold cup biopsies taken from the abnormal left upper pole renal calyx. 100x magnification demonstrating the presence of keratin (presumed to originate from an area of local squamous metaplasia) adjacent to normal mucosa. No clear dysplastic or malignant cells seen.

pelvis, only two cases describe the synchronous presence of a malignancy following histopathological examination of nephroureterectomy specimens; one transitional cell carcinoma and one squamous cell carcinoma.³ No features were identified that could relate the presence of KDSM to an increased risk of progression to malignancy.

Patients with SCI often require long-term indwelling catheters and are at increased risk of metaplastic changes in bladder urothelium. In a number of reported cases this has ultimately resulted in progression to invasive squamous cell carcinomas of the bladder, which tend to be of high grade.⁴

Currently there are no clear guidelines on the most appropriate management of KDSM in the upper

Figure 4 Images from same tissue sample as Figure 3, magnified to 200x.

urinary tract. Radical surgery was the mainstay of treatment for many years but, more recently, nephron-sparing approaches have advocated the combination of endoscopic and radiological surveillance. This report presents the first case of this rare diagnosis in a patient with SCI; a group of patients who inherently acquire risk factors for developing high-grade malignancy elsewhere in the urological tract. Preserving renal function in this case will require regular endoscopic keratin extraction, six-monthly surveillance with repeat biopsies, and serial CT imaging to ensure the disease neither progresses nor masks a more sinister underlying pathology.

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