Urothelial metaplasia of the seminal vesicle and ejaculatory duct associated with crossed-fused renal ectopia and Hutch diverticulum of the bladder

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The presence of urothelial epithelial metaplasia in a seminal vesicle is an exceptionally rare finding. We describe a unique case of urothelial metaplasia of the seminal vesicle and ejaculatory duct, found in a radical prostatectomy specimen from a patient with complex urogenital anatomy. A 70-year-old patient with organ confined (pT2) prostatic adenocarcinoma (Gleason score 3+4 = 7) had a right-sided Hutch diverticulum and a left crossed-fused renal ectopia. Although the histogenesis of urothelial metaplasia in the seminal vesicle remains unclear, in the patient presented herein it likely developed as a consequence of the previously unrecognized malformation.

Key Words: urothelial metaplasia, seminal vesicle, ejaculatory duct, Hutch diverticulum, crossed-fused renal ectopia

Introduction

Metaplasia is a reversible change, in which one differentiated cell type (epithelial or mesenchymal) is replaced by another mature cell type.[1] Although urothelial metaplasia is relatively common in the prostate, it is an exceptionally rare finding in the seminal vesicles, documented previously in only three case reports.[1-3] The histogenesis of urothelial metaplasia in the seminal vesicle is unclear, but it is believed to be a response to an adverse change in the native environment such as through mechanical irritation, inflammation, or infection.[2]

We describe a patient who had extensive urothelial metaplasia involving both the seminal vesicles (proximal and mid portion) and the ejaculatory ducts. He had a crossed-fused renal ectopia (CFRE), an anomaly where the kidneys are fused and located

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on the same side of the midline. The patient also had a Hutch bladder diverticulum, thought to be a rare abnormality due to congenital failure of normal muscle development around the ureteral orifice. Patients with Hutch diverticuli can be asymptomatic or may have diverse symptoms due to obstruction or voiding dysfunction, urinary retention inside the diverticulum (urinary infections) or stone formation.[4] Previous reports have associated Hutch diverticuli with vesicoureteral reflux and various degrees of renal dysmorphism,[4] but to our knowledge, urothelial metaplasia of the seminal vesicle and the ejaculatory duct has not been reported in this setting.

Case report

A 70-year-old man was referred for further evaluation after a nodule was identified on a digital rectal examination in the midline of the prostatic apex (cT2c), with a prostate specific antigen (PSA) value of 3.1 ng/mL. A 12-core transrectal ultrasound-guided biopsy revealed Gleason score 3+4 = 7 adenocarcinoma in 7 of 12 submitted cores (with 6 positive cores on the right side). The prostate volume was estimated at 29 cm^3, but no other preoperative imaging was performed.

The patient subsequently underwent a radical prostatectomy with the surgical pathology showing Gleason score 3+4 = 7 prostatic adenocarcinoma, with organ confined, node negative disease (pT2 pN0). Intraoperatively, the prostatectomy procedure was complicated by severe desmoplastic reaction at the right prostate base and the bladder neck. The right seminal vesicle was medially displaced, and after meticulous dissection, a large cystic structure was encountered posterior-lateral to the right bladder neck. The anterior bladder neck was opened to visualize the ureteric orifices and the right ureteric orifice

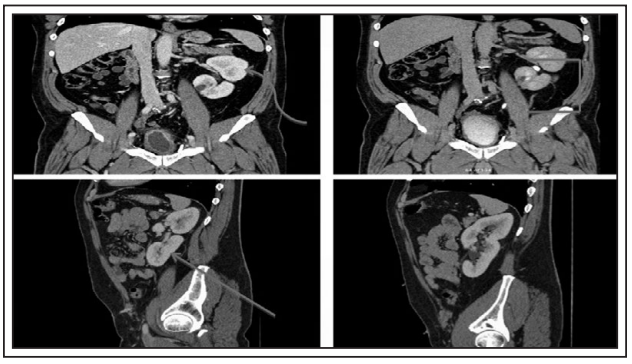

Figure 1. CT abdomen and pelvis enhanced. Curved arrow: left upper renal moiety; straight arrow: left lower renal moiety; bracket: stent in situ.

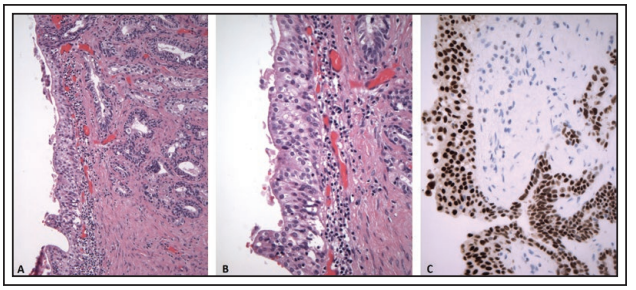

Figure 2. A) Luminal surface of the seminal vesicle is lined by unremarkable urothelium, indicating urothelial metaplasia. H&E stain (x 200); **B)** Higher magnification of A. H&E stain (x 400); **C)** GATA3 highlights the surface urothelial lining (x 400).

was followed into a large Hutch diverticulum. The diverticulum was repaired and the right ureter was re-implanted with placement of a right ureteric stent. Additional postoperative work up and imaging revealed a crossed-fused renal ectopia on the left side with the lower kidney's ureter crossing over to the right, Figure 1.

A 37 g prostate was submitted in-toto for pathologic evaluation, which revealed a metaplastic urothelial layer contiguously lining the seminal vesicles and the ejaculatory duct surface, and overlying the typical glandular structures, Figure 2A and 2B. Urothelial metaplasia was confirmed by the immunoreactivity for GATA-3, p63 and cytokeratin 7. The metaplastic seminal vesicle epithelium was cytokeratin 7 and GATA-3 positive, Figure 2C, but p63 only stained the basal cells. There was luminal ectasia, which extended along the ejaculatory ducts and periurethral glands, and the prostatic urethra was also dilated. There was no evidence of urothelial carcinoma in situ or invasive urothelial carcinoma. An abdominal postoperative ultrasound demonstrated presence of a CFRE, and was negative for any additional findings.

Two years following the radical prostatectomy, the patient is alive and without evidence of either disease recurrence or other complications. The patient's PSA was < 0.1 ug/L, he had a stable creatinine of 86 umol/L, with a negative urinalysis.

Discussion

We describe a unique case of urothelial metaplasia discovered in the setting of complex urogenital aberrations including CFRE and a Hutch diverticulum. The most prevalent manifestations of symptomatic Hutch diverticuli include urinary tract infections and ureteral obstruction due to narrowing of the ureter

at the neck of the diverticulum, however, neither symptom was present in our patient.[5] Hutch diverticuli have been associated with various degrees of renal dysmorphism including hypoplastic and absent kidney, although an association with CFRE has not yet been reported.[4,5] CFRE results from aberrant migration of the metanephric blastema and ureteral bud beyond the midline, and has a reported incidence on autopsy of 1:2000. It is typically asymptomatic without specific guidelines for management, and is usually detected incidentally on imaging studies, as in this case.[6]

Urothelial metaplasia of the seminal vesicle is also exceptionally rare and, to our knowledge, it has been documented only in two previous reports, with a total of three cases.[2,3] One report described two patients with radical prostatectomy specimens demonstrating an incidental finding of mature urothelium involving segments of the seminal vesicle. There was an abrupt transition to a normal simple cuboidal cell layer in one case, and circumferential urothelium was found in the other case.[2] The metaplastic changes were isolated to the seminal vesicles, and a clear etiology was not identified. Of note, the seminal vesicles and the bladder trigone are believed to develop as mesonephric (Wolffian) duct derivatives and are mesoderm-derived, which would suggest that seminal vesicle epithelium has potential to differentiate into urothelium.[3]

It was speculated that the development of urothelial metaplasia in the seminal vesicle may be a reaction to local mechanical irritation, inflammation, or infection.[2] This hypothesis is supported by one previous case of urothelial metaplasia in a seminal vesicle cyst, occurring in a setting of acute prostatitis and ejaculatory duct obstruction.[3] However, in the patient presented herein, urothelial metaplasia of the seminal vesicles and ejaculatory ducts occurred in the context of formerly unrecognized congenital malformations, including an unexpected intra-operative identification of a right-sided Hutch diverticulum, and a CFRE identified on postoperative imaging. As far as we are aware, the patient did not have a history of urinary tract infections or distal urinary obstructions, which may contribute or cause urothelial metaplasia of the seminal vesicles and ejaculatory ducts.

Urothelial metaplastic change in the seminal vesicle may potentially predispose to development of pre-neoplastic or neoplastic urothelial lesions, although no carcinoma in-situ or invasive urothelial carcinoma was identified in this case. Urothelial carcinoma involving the seminal vesicle appears to be a more common entity than urothelial metaplasia and has been reported arising either through mucosal spread or from the direct extension of muscle invasive urothelial carcinomas.[7,8] The most probable explanation for carcinoma in-situ of the seminal vesicle is intramucosal migration of the cancer in the prostatic ducts, and into the ejaculatory ducts and lumen of the seminal vesicles. Primary urothelial carcinoma of the seminal vesicles arising from metaplastic changes has yet to be reported.[8]

Urothelial metaplasia occurs much more commonly in the prostate than in the seminal vesicle. A study of urothelial and squamous metaplastic changes in prostatic tissue found no sign of a developing malignancy in these areas within a 2 year period.[1] Further research is required to determine whether these findings also hold true for metaplastic changes occurring in the seminal vesicles.

In summary, we describe a unique case of extensive urothelial metaplasia of the seminal vesicles and the ejaculatory ducts in a patient with complex urogenital malformations, consisting of Hutch diverticulum and CFRE, which has not been previously documented. Additional identification and documenting of urothelial metaplasia in the seminal vesicles is required to further study its possible pathogenesis and clinical significance. □

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