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VAGINAL STONE IN A MALE PATIENT WITH TRUE HERMAPHRODITISM

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A 31-year-old male with true hermaphroditism and a 46, XX karyotype who underwent gonadectomy and extirpation of the internal sex organs at the age of 4 had a large stone 4 cm in diameter in the residual male vagina. He complained of pain on micturition, hematuria, and rectal pressure. Urethroscopy and retrograde urethrography disclosed an ostium of the male vagina in the prostatic urethra, and an impacted intravaginal stone. Transurethral electrohydraulic lithotripsy was performed. Extracorporeal shock wave lithotripsy and transurethral lithotripsy were performed for the residual stones. All stones and fragments were spontaneously passed. The stone was composed of calcium phosphate and ammonium acid urate.

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Key words : Male vagina, Urolithiasis

INTRODUCTION

In males, secretion of the Müllerian regression factor by the fetal testes results in the presence of only vestigial female sex structures. The caudal end remains as the male vagina. An enlarged male vagina is often seen in association with proximal hypospadias or intersex disorders^{1,2)}. However, urolithiasis involving the male vagina is rare.

CASE REPORT

A 31-year-old male presented with pain on urination, gross hematuria, and rectal pressure. Urethroscopy and retrograde urethrography revealed a large ostium of the utricle in the prostatic urethra and an intrautricular stone 4 cm in diameter (Fig. 1). Computed tomography (CT) showed a calcified shadow between the urinary bladder and the rectum (Fig. 2).

When this patient was 4 years old, he was diagnosed with true hermaphroditism with a 46, XX karyotype. He underwent gonadectomy and extirpation of the internal sex organs. His chief complaint was gonadal dysfunction at his first visit to our hospital. He was given testosterone supplement therapy for 2 years.

Transurethral electrohydraulic lithotripsy (EHL) for the utricular stone was performed under spinal anesthesia on this admission. His penis was only 2 to 3 cm long. The urethral meatus was dilated with metal sounds. EHL was performed using a 9Fr probe through a cystoscope. The male vagina looked like a diverticulum of the urinary bladder. The stone had at least three layers, and the outer layer was very hard. The inner layers were comparatively fragile. For the residual stones, extracorporeal shock wave lithotripsy (ESWL) was performed three times with a Dornier MFL 5000 lithotripter. The patient

had no complaints while under epidural anesthesia. There was no gross hematuria. He voided the stone fragments. Finally, transurethral lithotripsy (TUL) was performed for the residual stones. These were also spontaneously passed. The stone was composed of calcium phosphate and ammonium acid urate.

DISCUSSION

The utricle is a rudimentary structure opening to the male prostatic urethra which is thought to be of mixed origin. The cranial portion is derived from the Müllerian duct, and the caudal segment is derived from the Wolffian and Müllerian ducts and the urogenital sinus³⁾. Enlarged prostatic utricles are seen in association with hypospadias in 11% to 14% of cases^{2,4)}. Based on urethrographic findings, prostatic utricles have been classified into four types: in grade 0, the opening is located on the posterior urethra, but the utricle does not extend over the verumontanum; in grade I, the opening is larger than in grade 0, but it does not reach the bladder neck; in grade II, which is more enlarged, the dome extends over the bladder neck; in grade III, the opening is situated in the bulbous urethra just distal to the external sphincter⁵⁾.

In the present case, the patient did not have hypospadias, and the utricle was grade 0. He was a true hermaphrodite with a 46, XX karyotype, mild gynecomastia, leg hair, and axillary hair. Furthermore, he had erectile and ejaculatory functions, although his penis was small. The origin of the stone was likely a lower urinary tract infection (UTI) due to a urethral stricture and residual urine in the male vagina. However, he had not been diagnosed with UTI previously.

This patient was mainly treated by ESWL, which has not been used for urethral stones⁶⁾. ESWL has

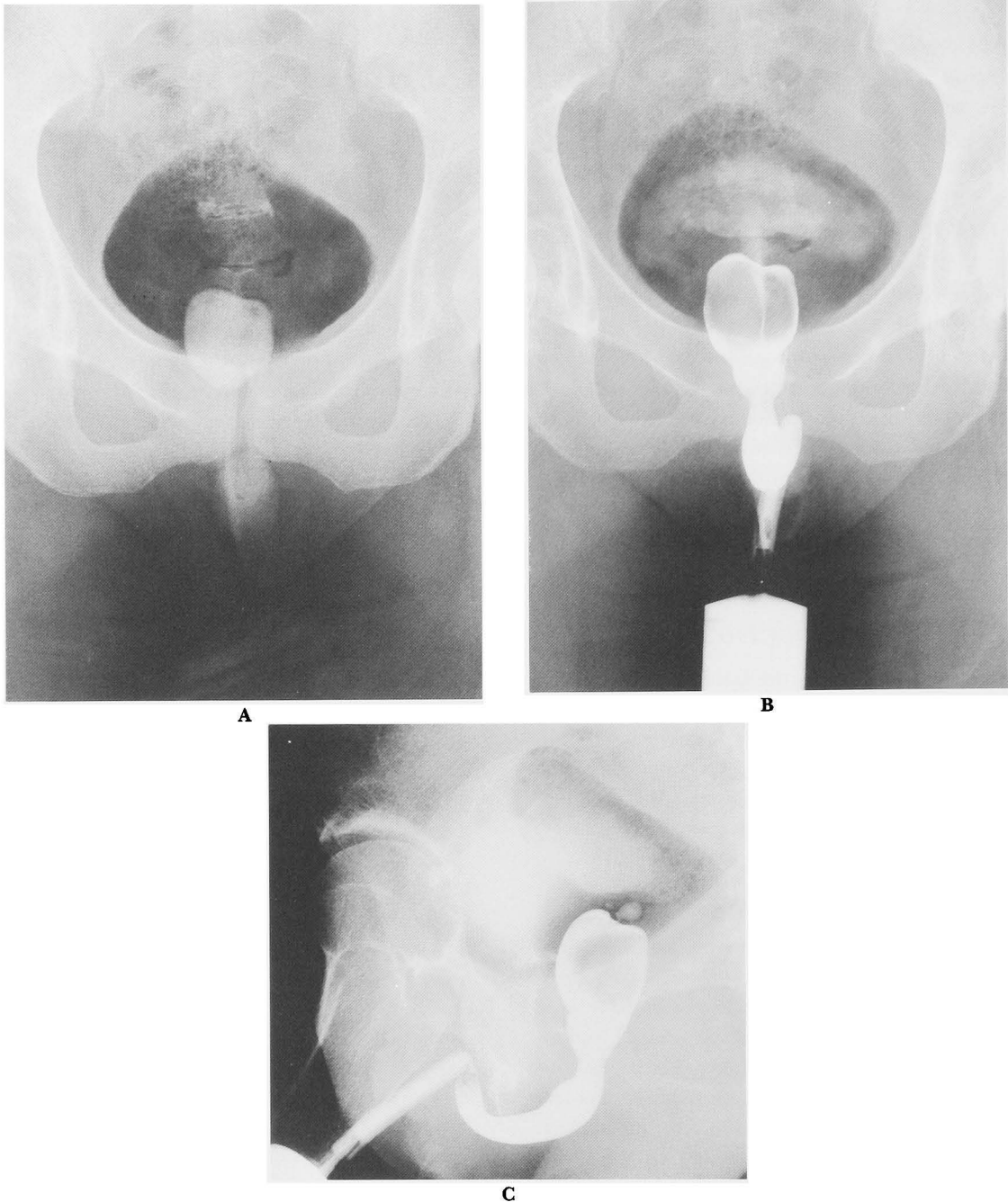


Fig. 1. Retrograde urethrography showing (A) a calcification in the pelvis, and (B, C) filling defects in the prostatic urethra.



Fig. 2. Computed tomography demonstrating a calcification between the urinary bladder and the rectum.

been successfully employed for a urethral stone in a boy with hypospadias after unsuccessful extraction maneuvers⁷⁾. Our patient refused subsequent endourology under anesthesia. Because EHL could not be continued, combination therapy with ESWL and TUL was performed. Male vaginal stones should be treated by EHL under endourologic guidance. ESWL is an alternative therapy.

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和文抄録

真性半陰陽に伴った男性子宮内に発症した結石の1例

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症例は31歳男性。染色体は46, XX。4歳時に子宮、性腺の摘出を受け、真性半陰陽と診断されている。排尿時痛、血尿を主訴として当科受診。尿道造影で後部尿道部に径4 cmの結石陰影を認め、尿道鏡にて男性子宮内結石と診断した。

経尿道的電気水圧式結石破碎術を行った。残石に対して体外衝撃波結石破碎術および経尿道的破碎術を行い完全に結石を破碎した。結石成分はリン酸カルシウムと酸性尿酸アンモニウムの混合石であった。

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