

Original Article

Giant prostatic utricular cyst with retrograde ejaculation: a case report and review of literature

Xiang-Hui Kong^{1,2}, Zhao-Hui Sun^{1,2}, Gang Chen^{1,2}, Wen-Jie Huang^{1,2}, Jie Zhang^{1,2}, Bo-Dong Lv^{1,2}, Xiao-Jun Huang^{1,2}

¹Department of Urology, The Second Affiliated Hospital of Zhejiang Chinese Medicine University, Hangzhou, Zhejiang Province, P. R. China; ²Zhejiang Provincial Key Laboratory of Traditional Chinese Medicine, Hangzhou, Zhejiang Province, P. R. China

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Abstract: The giant prostatic utricular cyst is a rare abnormality in the ejaculatory duct area. We report a 29-year-old male patient who had multiple semen examinations suggesting azoospermia. Transrectal ultrasonography (TRUS) showed a midline cyst of the prostate, about 6.0 cm in diameter. Pelvic magnetic resonance imaging (MRI) showed an approximately 7.0 cm diameter cyst between the seminal vesicles with high signal intensity on T1 and T2 weighted images. Urine sediment collected via prostate massage immediately after patient ejaculation contained a large amount of sperm. We performed a laparoscopic surgery for the prostatic utricular cyst, and successfully found sperm in semen postoperatively. The patient's symptoms, TRUS, MRI, and surgical results confirmed a diagnosis of giant prostatic utricular cyst with retrograde ejaculation. We review this case as well as previously reported cases and associated literature.

Keywords: Prostatic utricular cyst, retrograde ejaculation, infertility, laparoscopic cyst plastic surgery

Introduction

Midline cysts (MLCs) of the prostate are uncommon. With advances in radiological technology, such as transurethral ultrasound (TRUS) and magnetic resonance imaging (MRI), which can reveal cysts accurately, the incidence of MLCs has increased to 5-14%. Prostatic utricular cyst is the most common cyst in this region. The development process of MLCs is unclear, but Müllerian duct and prostatic utricular cysts may be related to deficiencies of testosterone and Müllerian inhibitory substance during embryonic development [1]. Although 95% of these cysts are asymptomatic, other people may suffer urinary tract infections, obstructive or irritative voiding symptoms, chronic pelvic pain syndrome, epididymitis, hematospermia, low semen volume or even infertility [2]. However, recent literature has not reported retrograde ejaculation caused by a huge prostatic utricular cyst. Retrograde ejaculation (RE) occurs when the semen does not exit the urethra but instead enters the bladder due to the weakness of the corpus cavernosum and the ischial corpus cav-

ernosum. We suggest that giant prostatic utricular cysts may cause this unique type of RE.

Case report

The patient, a 29-year-old male, was first admitted to the Second Affiliated Hospital of Zhejiang University School of Medicine (Zhejiang, China) for infertility and a prostate cyst, without erectile dysfunction, ejaculation pain, dysuria, or hematuria. In the past 3 years, the patient's ejaculation and climax feelings occurred in every sexual encounter, but the amount of ejaculate was low, at only about 1.5 ml. The semen analysis indicated azoospermia.

On physical examination, the vas deferens was felt on the left, but not on the right. We excluded genitalia, testis and secondary sexual characteristic problems. Digital rectal examination revealed an obvious cystic enlargement in the midline of the prostate which was smooth and elastic. Biochemical and sex hormones were normal. Karyotype analysis was normal, with XY sex chromosomes. TRUS revealed a midline

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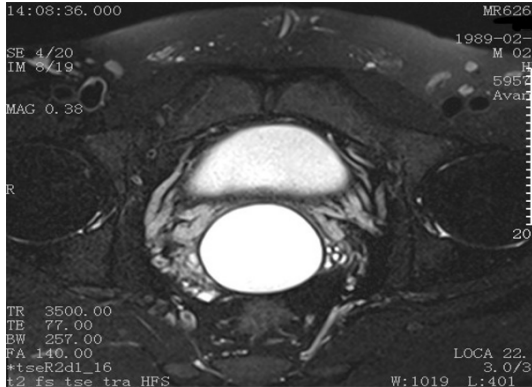


Figure 1. The MRI image of the seminal vesicles (SV) and prostate. There is a 7.0*5.4 cm high signal intensity cyst in the midline region of the prostate on T2 weighted images.

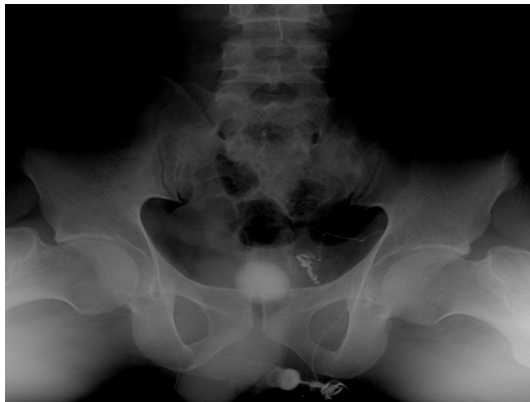


Figure 2. Pelvic radiography. The left vas deferens and seminal vesicle are well developed, and a high density area is seen in the midline of the prostate.

prostatic cyst, about 6.0 cm in diameter. Pelvic MRI confirmed a 7.0 cm diameter high signal intensity cyst in the midline region of the prostate on T1 and T2 weighted images (**Figure 1**). After ejaculating, the patient underwent immediate prostate massage. The urine examination revealed a sperm concentration of $126.29 \times 10^6/\text{ml}$, with 6% motile sperm and 94% inactive sperm.

In order to determine the location of the cyst, vasography was performed. During the operation, there was no vas deferens in the tail of the epididymis, and the right vas deferens could not be found. We separated the left vas deferens and punctured it, then injected it with meglumine diatrizoate. Radiography showed that the left vas deferens and seminal vesicle were well developed, and a high density area

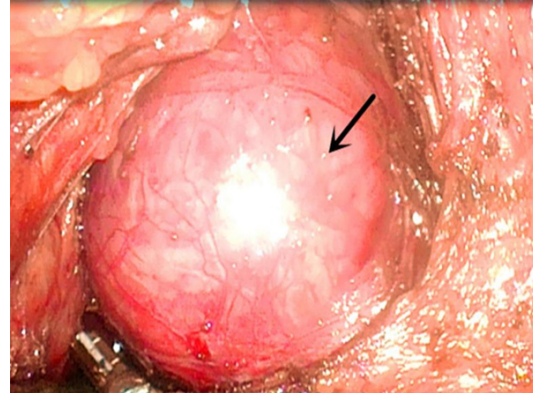


Figure 3. A cyst located between the bladder and rectum, approximately 7.0 cm in diameter.

could be seen in the midline of the prostate (**Figure 2**). Seminal vesiculoscopy indicated that there was no obstruction or stones in the urethra and the prostatic utricle opened on the verumontanum. After entering the prostatic utricle through the orifice, we found that the cavity was huge, the fluid was turbid, and there were no stones or tumors. Methylthionium chloride injected into the left vas deferens was seen to be ejected out of the lower side of the cyst.

Further laparoscopic plastic surgery was continued. Incision of the peritoneum in the posterior part of the bladder revealed that the prostatic utricular cyst was about 7 cm in diameter, located between the bladder and rectum (**Figure 3**). The vas deferens and seminal vesicle could be seen on both sides of the cyst. The right vas deferens was hypoplastic, becoming thinner near the orifice of the abdominal ring and disappearing gradually. The cyst was fully separated and opened, and contained a large amount of yellowish-brown fluid. The left ejaculatory duct was connected with the cyst (**Figure 4**), but the right was not, and there was an approximately 0.3 cm diameter orifice between the cyst and the urethra. A guide wire inserted from the cyst through the urethra confirmed that the cyst was connected with the urethra (**Figure 5**). We resected the superfluous wall of the cyst and part of the right vas deferens and seminal vesicle for pathological examination. The prostatic utricular cyst was trimmed to establish a tubular channel from the orifice of the left vas deferens to the urethra. The double J stent tube and the catheter were retained.

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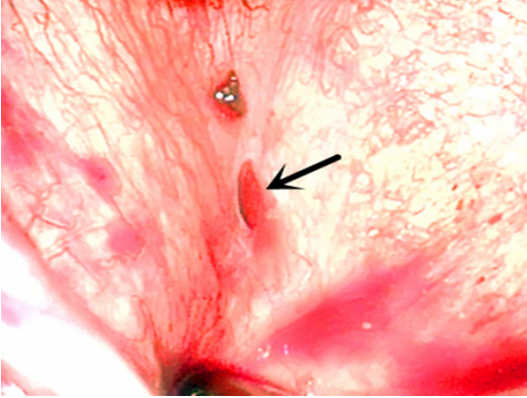


Figure 4. The orifice of the left ejaculatory duct is located in the cyst.

Postoperative pathology showed that the prostatic utricular cyst and the right vas deferens near the deep ring of the inguinal canal were almost occluded. One month after operation, a routine semen exam showed that semen volume was 2.3 ml and sperm concentration was $0.6 \times 10^6/\text{ml}$. Therefore, it is considered that the existence of the giant prostatic utricular cyst resulted in a unique type of retrograde ejaculation and secondary infertility.

Discussion

Reproductive system cystic lesions mainly include prostatic utricle, Müllerian, ejaculatory duct and seminal vesicle cysts. MLCs are mostly asymptomatic. Only 5% of MLCs are associated with chronic pelvic pain syndrome, hematospermia, painful ejaculation, voiding dysfunction, prostatitis-like syndrome, and infertility. Prostatic utricle cysts are the most common of these cystic lesions [2, 3]. The Müllerian duct plays an important role in the occurrence of prostate utricle cyst and Müllerian cysts. This duct usually develops as the fallopian tubes, uterus and vagina in females. However, in some males there is no degradation of the Müllerian ducts during fetal development, which can lead to abnormal expansion of the prostate utricle and the formation of Müllerian cysts [4]. The prostatic utricle is a small diverticulum in the male prostatic urethra. It is located on the top of the colliculus seminalis and in the middle of the two ejaculatory duct orifices. After abnormal enlargement, a prostatic utricular cyst is produced. The cyst communicates with the urethra and the ejaculatory duct. It is easily con-

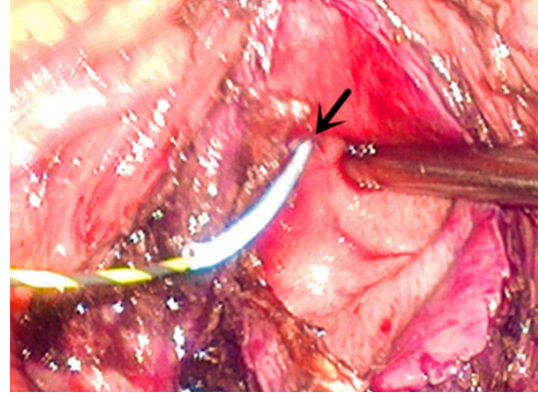


Figure 5. An approximately 0.3 cm diameter orifice between the cyst and urethra. A guide wire is inserted from the cyst in the direction of urethra.

fused with Müllerian cysts. The Müllerian cyst is located in the midline of the prostate but does not communicate with the ejaculatory duct, urethra and seminal vesicles [5]. The ejaculatory duct cyst is located paramedially along the ejaculatory duct and is usually caused by a unilateral or bilateral cystic dilatation of the ejaculatory duct. The seminal vesicle cyst is cystic dilatation of the unilateral or bilateral seminal vesicle, along with the disappearance of convolutions of tubules and ipsilateral renal anomalies [3].

TRUS can indicate the presence of a midline prostate cyst and the condition of the prostate. Because TRUS is non-invasive, inexpensive, and trustworthy, it has become the first choice for the diagnosis of cystic lesions of the genitourinary tract. However, diagnosis via TRUS is subjective and closely related to the experience level of the physician. MRI can be used as a gold standard for the diagnosis of various cyst diseases. It is easier to visualize lesions, and can be used for patients with difficult diagnosis by TRUS [6-8].

This patient had a large cyst, 7 cm in diameter, located in the posterior superior margin of the prostate. It was easily considered to be a Müllerian cyst on MRI. However, MRI did not show the laterally curved ejaculatory duct, nor could we determine the communication between the cyst and the urethra. Therefore, we performed vas deferens angiography and seminal vesiculoscopy [9, 10]. We found that the cyst had a channel communicating with the urethra, the ejaculatory duct was abnormally open in

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the cyst, and the seminal vesiculoscope could be inserted into the cyst. In consideration of these facts, we diagnosed a prostatic utricular cyst.

Retrograde ejaculation is usually due to the weakness of the corpus cavernosum and the ischial corpus cavernosum during ejaculation, so that the semen is partially or completely unable to be ejected to the urethra, but reverses into the bladder. RE is a cause of oligospermia or azoospermia, and eventually leads to infertility and loss of orgasm. Any patient with semen volume less than 2 ml should be suspected of having RE [11]. Urine sediment examination can aid in diagnosis, as it will show the presence of sperm after retrograde ejaculation. In this case, the patient's semen examination showed no sperm, and the amount of semen in the ejaculate was only about 1.5 ml. However, examination of the urine sediment obtained by prostate massage immediately after ejaculation showed a large amount of sperm. Since the ejaculatory duct opened into the cyst, the semen flowed into the cyst during ejaculation and nothing or only a small amount sperm could be directly injected through the urethra. During prostate massage, the external force caused some of the sperm that was retained in the cyst to overflow through the posterior urethra. Therefore, we found a large amount of sperm in the microscopic examination of the urine sediment. The majority of the sperm was non-viable, and we speculate that the semen had accumulated in the cyst after the previous ejaculation. Over time, the sperm lost vitality due to the lack of growth environment. In summary, we believe that huge prostatic utricular cysts can cause retrograde ejaculation.

Several studies have assessed MLCs in infertile or otherwise symptomatic patients. Lotti et al, discovered that MLCs were present in 66/648 (10.2%) of infertile males, and patients with MLCs are more likely to be infertile. They also believe if the volume of the midline prostate cyst is less than 0.117 ml, the impact on semen is extremely low, and further treatment is unnecessary [12]. Kang found MLCs in 61 symptomatic cases, and the mean diameter of the cysts was 0.99 cm (range 0.5-3.0 cm) [9]. Li observed 4 cases of large cystic structures ranging from 3.5 × 4.4 cm to 8.0 × 10.5 cm in size, but the further treatment and fertility of these cases was not reported [7]. Some researchers claim that treatment should only

be performed on symptomatic or infertile patients. Treatments such as transrectal puncture, transurethral unroofing via resectoscope, open surgery, and laparoscopic cyst surgery can improve the semen quality. Transrectal puncture and transurethral unroofing via resectoscope have been identified to improve patients' oligospermia and azoospermia, and even help patients to conceive successfully after surgery [10, 12].

In our patient, the 7.0 cm diameter cyst that had caused symptoms similar to retrograde ejaculation was considered too large for effective treatment with transrectal puncture or transurethral unroofing via resectoscope. Laparoscopic cyst surgery allowed visible resection of the superfluous cyst wall, and reduced the chance of rectal injury. The remaining cyst wall was sutured to form a tubular channel, through which the orifice of the ejaculatory duct was connected with the opening of the prostate utricle, so that the semen can directly exit through the tubular passage and the urethra to reduce semen retention and effectively improve the postoperative semen quality.

In summary, a large prostatic utricular cyst can cause a unique type of retrograde ejaculation. When retrograde ejaculation occurs with these cysts, laparoscopic cyst surgery is recommended to improve the patient's symptoms and to increase the chance of natural conception.

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Disclosure of conflict of interest

None.

Address correspondence to: Dr. Xiao-Jun Huang, Department of Urology, The Second Affiliated Hospital of Zhejiang Chinese Medicine University. No. 318, Chao Wang Road, Hangzhou 310005, Zhejiang Province, P. R. China. Tel: 86+0571-85288-276; Fax: 86+0571-88064725; E-mail: hxj19682-002@126.com

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