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Prostate Cystadenoma Presenting With Obstructive Azoospermia

Case Report

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ABSTRACT: We report on a case of prostate multilocular cystadenoma presenting with primary infertility and obstructive azoospermia. Our patient is a 36-year-old presenting with primary infertility in addition to mild deep pelvic pain. Semen analysis revealed azoospermia with positive fructose. His prostate-specific antigen was 0.7 ng/dL and his imaging revealed a large multilocular cystic mass with multiple internal enhancing septa. Transrectal ultrasonography-guided aspiration and biopsies revealed a lining of regular low cuboidal cells. Surgical removal was undertaken through a transperitoneal/retroperitoneal approach and pathology was consistent with a prostatic multilocular cystadenoma. Further studies are needed to characterize and classify cystic lesions of the prostate.

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Multilocular cystadenoma of the prostate is a rare tumor, with only 14 cases reported in the literature (Park et al, 2007; Tuziak et al, 2007; Chowdhury and Abdulkarim, 2009). It usually presents clinically as a multiseptate large cystic pelvic mass, with associated symptoms including lower urinary tract symptoms (LUTS), urinary retention, and pain (Galosi et al, 2009). We report on a case of prostate multilocular cystadenoma presenting with primary infertility and obstructive azoospermia.

Case Report

Our case is a 36-year-old patient presenting with primary infertility. He was evaluated by complete history and physical examination, semen analysis, prostate-specific antigen (PSA), and imaging by transrectal ultrasonography (TRUS) and magnetic resonance imaging (MRI) with TRUS-guided biopsies, and finally the multilocular cystadenoma was resected through a lower midline transperitoneal/retroperitoneal approach. The study was approved by the institutional review board.

Results

The patient presented with primary infertility. He also complained of mild deep pelvic pain and perineal discomfort for more than 10 years. His semen analysis revealed azoospermia with positive fructose. His scrotal examination revealed the testes to be normal in size and consistent with bilaterally palpable vasa deferentia and no epididymal or spermatic cord abnormality. The patient's serum testosterone and follicle-stimulating hormone were within normal. His PSA was 0.7 ng/dL, and imaging with TRUS and pelvic MRI (Figures 1 and 2) revealed a large multilocular cystic mass ($9.0 \times 7.0 \times 6.0$ cm) in the pelvic cavity encircling and stretching the urethra with multiple internal enhancing septa. The mass displaced the urinary bladder and seminal vesicles superiorly and anteriorly with no evidence of invasion, being posteriorly related to the rectosigmoid with preserved cleavage plane. It extended caudally to the level of the origin of penile crura.

TRUS-guided aspiration and biopsies revealed the lining of the septa to be formed of regular low cuboidal cells with no evidence of malignancy. Surgical removal of the mass (partial prostatectomy) was undertaken through a transperitoneal/retroperitoneal approach. The final pathology was consistent with a prostatic multilocular cystadenoma (Figures 3 and 4).

The postoperative period was marked by the occurrence of significant urinary extravasation from the posterior urethra upon removal of the urethral catheter on the seventh postoperative day, necessitating replacement of the catheter and prolonged indwelling drainage for 5 more weeks. The catheter was eventually removed after a cystourethrogram confirming absence of urinary leak. One month following catheter removal, the patient was voiding satisfactorily with no LUTS and no pelvic pain. He had resumed sexual activity and reported excellent potency; however, he noted a marked decrease

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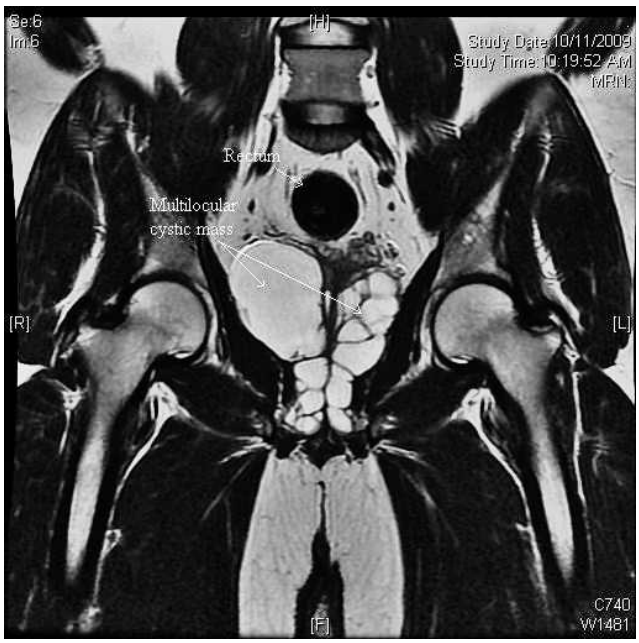

Figure 1. Coronal T_2-weighted scan.

in semen volume. A postcoital urine analysis showed numerous sperm and confirmed the diagnosis of retrograde ejaculation. The patient finally elected to proceed with testicular biopsy and testicular sperm extraction, which revealed normal spermatogenesis, and sperm were retrieved and cryopreserved for in vitro fertilization with intracytoplasmic sperm injection.

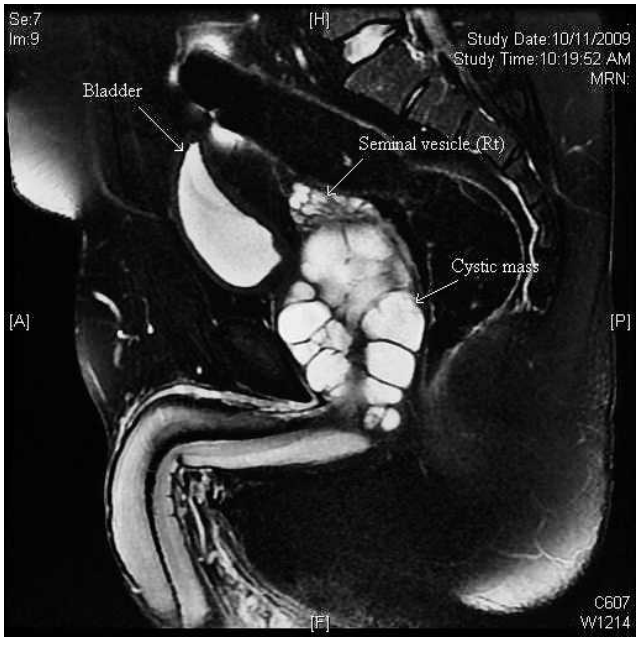

Figure 2. Sagittal T_2-weighted scan.

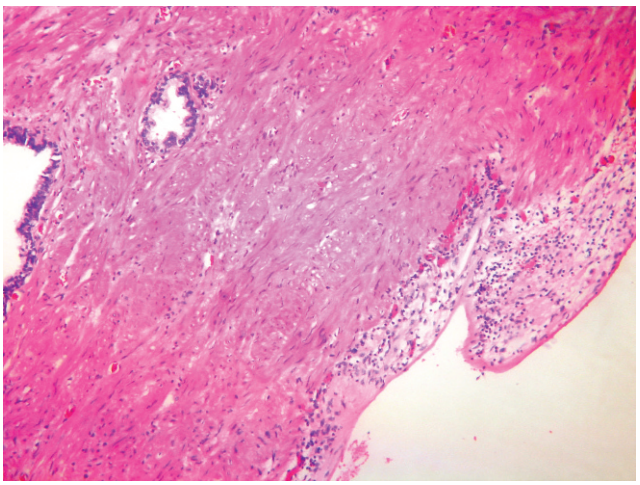

Figure 3. Wall of prostate cystadenoma with focal ulceration (×10). Color figure available online at www.androlgyjournal.org.

Discussion

Prostatic cystadenoma is a rare prostatic lesion, with only a few cases reported in the literature during the last 3 decades. The term ''giant multilocular prostatic cystadenoma'' was proposed by Maluf et al (1991) to describe these large multiseptate cystic pelvic masses with dimensions ranging between 9 and 45 cm. The tumor can be observed during TRUS, computed tomography, or MRI to have solid and anechoic content, with thick and irregular walls or hairline septa (Galosi et al, 2009). The origin from the prostate may or may not be appreciated, as the mass can extend into extraprostatic tissues and then into the pelvic or suprapubic space.

Symptoms described in association with the mass have included LUTS, urinary retention, pain, and

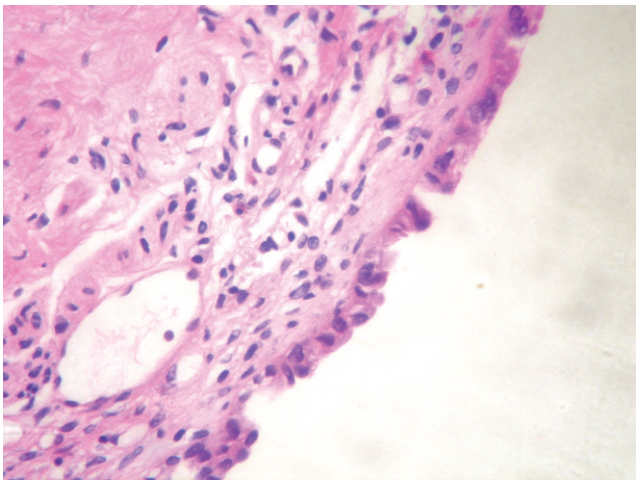

Figure 4. Low cuboidal lining with mild inflammation (×20). Color figure available online at www.androlgyjournal.org.

hematuria. To our knowledge our patient is the first to present with primary infertility and the second to have obstructive azoospermia (Park et al, 2007). The suggested cause of obstructive aspermia in that report was fibrous obliteration of portions of the vas deferens and seminal vesicles. Serum PSA could be normal or increased.

Histologically, epithelial cells have distinctive features. A single layer of cuboidal cells lines the cystic spaces, and peribasal cells are present with nuclei showing no atypia or prominent nucleoli.

The growth pattern of cystadenoma is expansile and not invasive; however, adherence to adjacent organs was reported (Choi et al, 2000) and so was recurrence following incomplete resection (Rusch et al, 2002). Our surgery was performed through a combined retrovesical and retropubic approach. The main challenge during surgery was to completely resect the cystic tumor while preserving the seminal vesicles, vasa deferentia, and neurovascular bundles. A significant part of operative time was spent identifying these structures and carefully dissecting them away from the lesion to be resected. The postoperative period was marked with significant urinary extravasation from the posterior urethra following removal of the indwelling urethral catheter on the seventh postoperative day, requiring replacement of the catheter and prolonged catheter drainage for 6 weeks. Following recovery, the patient maintained his potency, but the surgery resulted in retrograde ejaculation and the patient went on to testicular sperm extraction and cryopreservation. We believe thorough preoperative counseling and continued postoperative support helped the patient cope with the prolonged convalescence and final outcome.

Prostate multilocular cystadenoma is a rare tumor that can present with primary infertility and obstructive azoospermia. Surgical resection of the lesion may have significant impact on sexual function and fertility. Further studies are needed to characterize and classify this entity and other cystic lesions of the prostate.

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