Case Report

Robot-assisted laparoscopic vesicule prostatectomy for mixed epithelial–stromal tumor of seminal vesicle

Yuki Masuo,1 Hisanori Taniguchi,1 Tomoaki Matsuzaki,1 Hidefumi Kinoshita,1 Chika Miyasaka,2 Chisato Ohe2 and Tadashi Matsuda1

Departments of 1Urology and Andrology, and 2Pathology and Laboratory Medicine, Kansai Medical University, Hirakata, Osaka, Japan

Abbreviations & Acronyms
CT = computed tomography
HE = hematoxylin and eosin
IQR = interquartile range
LUTS = lower urinary tract symptoms
MEST = mixed epithelial–stromal tumor
MS = metastasis
NA = not available
NED = no evidence of disease
PSA = prostate-specific antigen
RALVP = robot-assisted laparoscopic vesicule prostatectomy
S-DRE = screening digital rectal examination
SV = seminal vesicle

Introduction: Mixed epithelial–stromal tumor is a biphasic tumor with stromal and benign epithelial components. Only 40 cases of mixed epithelial-stromal tumor originating from a seminal vesicle have previously been published in English.

Case presentation: A 52-year-old man was transferred to our hospital for evaluation of a 3.0-cm pelvic tumor detected incidentally by computed tomography. Robot-assisted laparoscopic vesicule prostatectomy was performed. We approached the Retzius space from both levels of the pouch of Douglas and peritoneal top of the bladder to clarify the tumor's environment. Pathologically, the tumor was diagnosed as a low-grade mixed epithelial–stromal tumor originating from the right seminal vesicle. There was no evidence of disease recurrence within 51 months.

Conclusion: This is the first report of robot-assisted laparoscopic vesicule prostatectomy for a seminal vesicle mixed epithelial–stromal tumor. Long-term observation is warranted due to the lack of reports with sufficient follow-up to ensure the procedure’s safety.

Key words: mixed epithelial–stromal tumor, prostate, robot-assisted laparoscopic prostatectomy, robot-assisted laparoscopic vesicule prostatectomy, seminal vesicle.

Keynote message
A 52-year-old man with a MEST originated from the SV underwent RALVP. This is the first case of RALVP performed for MEST of a SV. We report on the surgical technique and pathological findings of the current and previous reported cases of MEST arising from a SV.

Primary SV tumors are rare. The most common malignant tumor of the SV is adenocarcinoma, followed by sarcoma and tumors with mixed epithelial and stromal components.1 MEST is a biphasic tumor with stromal and benign epithelial components that was referred to by various terms until the most recent edition of the World Health Organization classification in 2016. However, SV MESTs have rarely been reported.1 We herein report a patient with MEST originating from the right SV who underwent RALVP.

Case presentation
A 52-year-old asymptomatic Asian man was transferred to our hospital for evaluation of a pelvic tumor that was detected incidentally by CT following colon cancer surgery 3 years earlier. Magnetic resonance imaging confirmed a 3.0 × 3.0 × 3.2 cm mass in the middle of the SV, with a thin capsule with contrast-enhanced irregularities of low and high signal intensities by T1- and T2-weighted imaging, respectively (Fig. 1a). The tumor was indistinct from the SVs and prostate with no local extension or lymphadenopathy. His serum PSA level was 0.47 ng/mL. Transrectal needle biopsy of the tumor was performed and pathological examination indicated a spindle cell neoplasm, suggesting a possible stromal tumor of uncertain malignant potential on the prostate.
RALVP including tumor resection with bilateral nerve preservation was performed using a four-arm Da Vinci Si system. During surgery, we initially approached the Douglas pouch to clarify the tumor’s environment. After transverse incision of the peritoneum at the level of the pouch of Douglas, the surrounding tissues were carefully released. The peritoneal top of the bladder was then incised again to approach the Retzius space. The tumor became apparent after separating the prostate from the bladder neck. Bilateral neurovascular bundles were spared using the intrafascial approach.2

There were no intraoperative or postoperative complications. The patient was discharged on postoperative day 8 with normal voiding. There was no CT evidence of disease recurrence within 30 months. The patient wears an occasional pad for safety, but his erectile function has returned and sexual intercourse is possible without phosphodiesterase inhibitors.

**Surgical specimen and histopathology**

Macroscopically, the tumor was well circumscribed and arose from the right SV. The prostate was clearly separated from...
the tumor. A cross section showed a solid tan–white mass centered in the region of the SV. No gross areas of hemorrhage or coagulative necrosis were seen (Fig. 1a–d).

Microscopic findings revealed stromal and epithelial tumor components. The stromal component comprised spindle cells with varying degrees of cellularity. Mild nuclear atypia and pleomorphism were focally present (Fig. 2a–d). These cells showed no evident mitotic activity (<1/10 high-power field). The epithelial component comprised dilated, large, lined cuboidal epithelial cells.

Immunohistochemically, spindle cells in the stromal component were positive for CD34, estrogen receptor, progesterone receptor, and desmin, but negative for Ki-67 (<1%) and p53 (Fig. 3). The stromal component was positive for AE-1/3 but negative for PSA and prostatic acid phosphatase. Based on these findings, the pathological diagnosis was low-grade MEST originating from a SV.

Discussion

We report on a middle-aged man who underwent RALVP for MEST originating from the right SV.

According to the most recent edition of the World Health Organization’s classification of Tumors, Pathology and Genetics, MEST including neoplasms previously called “cystadenoma,” “epithelial–stromal tumor,” “cystomyoma,” “cystic epithelial–stromal tumor,” and “mesenchymoma.” These tumors were defined as “MEST which are biphasic tumors with stromal and benign epithelial components.” Pathologically, MEST is classified as low, intermediate, or high grade. Reikie et al. proposed a distinction of grade based on the histologic characteristics including stromal atypia, mitotic activity, nuclear pleomorphism, and tumor necrosis.

An English-language PubMed search including 24 reports reviewed by Reikie et al. and the current case identified 41 cases of MEST arising from a SV. Excluding one case in which the tumor was detected at autopsy, 41 cases reported since 1944 are summarized in Table 1.

The median age of the 41 patients was 49.0 years (IQR 43.0–59.0 years). Many cases were diagnosed as cystadenoma. The median tumor diameter in 39 cases (two cases did not supply the tumor size) was 7.5 cm (IQR 5.5–12.0 cm). The surgical approach depended on the anatomic lesion, tumor size, and surgeon’s expertise. Robot-assisted laparoscopic surgery was performed in recent cases.

The median duration of follow-up for 32 cases after their first surgical approach was 21.0 months (IQR 11.75–39.0 months). The outcome in most cases was “NED.” However, two cases had local recurrence diagnosed pathologically as low and intermediate grade, respectively, and another two had lung metastases within 48 months, diagnosed as high grade. One patient with high-grade disease died 11 months after metastatectomy. These findings suggest that high-grade MEST requires strict follow-up after treatment.

The current patient was 52 years old and relatively small size. RALVP including complete tumor resection with bilateral nerve preservation was performed. The patient remained alive with NED recurrence 51 months after surgery. Lober et al. reported a patient with a low-grade tumor who was asymptomatic at the time of diagnosis; however, the tumor increased five-fold in volume and became symptomatic 10 years later, when surgical removal of the mass was much more difficult. Bullock also reported a 12-cm low-grade tumor with local recurrence 36 months after treatment due to incomplete resection. These cases suggest that the strategy in the current case was appropriate.

Conclusion

This is the first report of RALVP performed for a MEST of the SV. Long-term observation is warranted because of a lack of follow-up evidence to ensure the procedure’s safety.
| Author          | Year | Age (years) | Author’s terminology | Size (cm) | Symptom | Surgical approach | Grade | Follow-up (months) | Outcome         |
|-----------------|------|-------------|----------------------|-----------|---------|-------------------|-------|-------------------|----------------|
| Plaut et al.    | 1944 | 66          | Cystomyoma           | 15        | +       | Palpable abdominal mass | Tumorectomy | Low               | 5 NED           |
| Soule et al.    | 1951 | 47          | Cystadenoma          | 14 +      | LUTS, fatigue | NA               | Low  | 300               | NED             |
| Islam et al.    | 1979 | 37          | Mesenchymoma         | 5.5       | –       | S-DRE              | Low   | 60                | NED             |
| Lundhus et al.  | 1984 | 39          | Cystadenoma          | 9         | +       | LUTS, abdominal/ perineal pain | Vesiculectomy | Low  | 3                | NED             |
| Mazur et al.    | 1987 | 49          | Cystic epithelial stromal tumor | 7       | +       | LUTS              | Tumorectomy | Intermediate | 24 Recurrence, 18 months after final resection |
| Bullock et al.  | 1988 | 59          | Cystadenoma          | 12        | +       | LUTS              | Vesiculectomy | Low  | 36               | Recurrence, 36 months after final resection |
| Raghuveer et al.| 1989 | 45          | Cystadenoma          | 5.3       | +       | LUTS, abdominal pain | Tumorectomy | Low  | 16               | NED             |
| Mazzucchelli et al.| 1992 | 63          | Cystadenoma          | 3         | +       | Inguinal pain      | Vesiculectomy | Low  | 96               | NED             |
| Laurila et al.  | 1992 | 49          | Mullerian adenosarcoma-like tumor | 6       | +       | LUTS, palpable abdominal mass | Cystoprostatectomy | Intermediate | 48 NED          |
| Ranschaert et al.| 1992 | 50          | Cystadenoma          | 12        | +       | LUTS              | Visculectomy | Low  | –                | NA              |
| Lagalla et al.  | 1993 | 33          | Cystadenoma          | NA        | +       | Hematospermia, hematuria | Tumorectomy | Low  | –                | NA              |
| Fain et al.     | 1993 | 61          | Cystosarcoma phylloides | 8.5      | +       | LUTS              | Cystoprostatectomy | High | 48               | Lung M5, 6 months after chemotherapy NED |
| Peker et al.    | 1997 | 47          | Cystadenoma          | 8         | +       | Hematospermia, suprapubic discomfort, tenesmus | Visculectomy | Low  | 21               | NED             |
| Baschinsky et al.| 1998 | 37          | Cystadenoma          | 6.5       | +       | LUTS, hematospermia | Cystoprostatectomy | Low  | 6                | NED             |
| Santos et al.   | 2001 | 49          | Cystadenoma          | 16        | +       | LUTS              | Tumorectomy | Low  | 27               | NED             |
| Abe et al.      | 2002 | 65          | Cystosarcoma phylloides | 6       | +       | LUTS              | Visculectomy | High | 11               | Lung M5, died 11 months after resection |
| Gil et al.      | 2003 | 49          | Phylloides tumor     | 7         | –       | S-DRE              | Tumorectomy | Low  | 36               | NED             |
| Son et al.      | 2004 | 38          | Phylloides tumor     | 16        | +       | LUTS, abdominal pain | Visculectomy | Intermediate | 24 NED          |
| Lee et al.      | 2006 | 46          | Cystadenoma          | 7.5       | –       | S-DRE              | Visculectomy | Low  | 6                | NED             |
| Hoshi et al.    | 2006 | 70          | Epithelial stromal tumor | 4.6      | +       | Abdominal pain, fatigue | Cystoprostatectomy | Low  | 14               | NED             |
| Khan et al.     | 2007 | 43          | Phylloides tumor     | 5.5       | +       | Hematospermia, testicular pain | Visculectomy | Low  | 1                | NED             |
| Monica et al.   | 2008 | 50          | Epithelial stromal tumor | 9       | +       | LUTS, fever       | Tumorectomy | Low  | 26               | NED             |
| Thway et al.    | 2008 | 61          | Epithelial stromal tumor | 8       | +       | Hematospermia, suprapubic pain, tenesmus | Visculectomy | Low  | 21               | NED             |
| Lorber et al.   | 2011 | 52          | Cystadenoma          | 14        | +       | LUTS              | Visculectomy | Low  | –                | NA              |
| Ploumidis et al.| 2012 | 45          | Cystadenoma          | 17.2      | +       | LUTS, pelvic pain  | Visculectomy | Low  | –                | NA              |
| Zhu et al.      | 2013 | 31          | Cystadenoma          | 8.8       | +       | Hematospermia     | Visculectomy | Low  | –                | NA              |
| Arora et al.    | 2013 | 23          | Cystadenoma          | NA        | +       | LUTS, abdominal pain | Tumorectomy | Low  | –                | NA              |
| Zhang et al.    | 2013 | 32          | Cystadenoma          | 5         | +       | Hematospermia     | Visculectomy | Low  | 19               | NED             |
| Zhang et al.    | 2013 | 64          | Cystadenoma          | 4.5       | +       | Perineal pain     | Visculectomy | Low  | 82               | NED             |
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Conflict of interest

The authors declare no conflict of interest.

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