Case Report

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

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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

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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 vesicle 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 vesicle 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 vesicle 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.

Introduction

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.

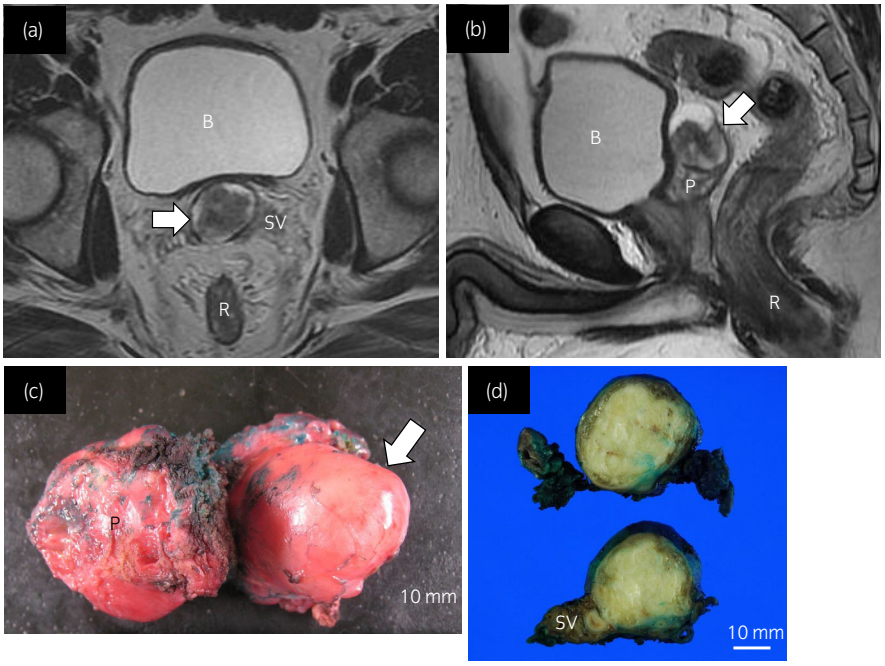

Fig. 1 Pelvic magnetic resonance imaging showed a 3.0 × 3.0 × 3.2 cm solid tumor in the middle of the two SVs with a thin capsule of low-signal intensity (arrows). The mass was indistinct from the SVs and prostate. Surgical specimen resected by RALVP. (a) T2-weighted coronal image, (b) T2-weighted sagittal image. (c) Macroscopically, the tumor was 3.0 × 3.0 × 3.2 cm in diameter and well circumscribed, and arose from the right SV (arrow). The prostate was clearly separated from the tumor. (d) Cross section of a solid tan–white mass centered in the region of the SV. No gross areas of hemorrhage or coagulative necrosis were seen. B, bladder; P, prostate; R, rectum.

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

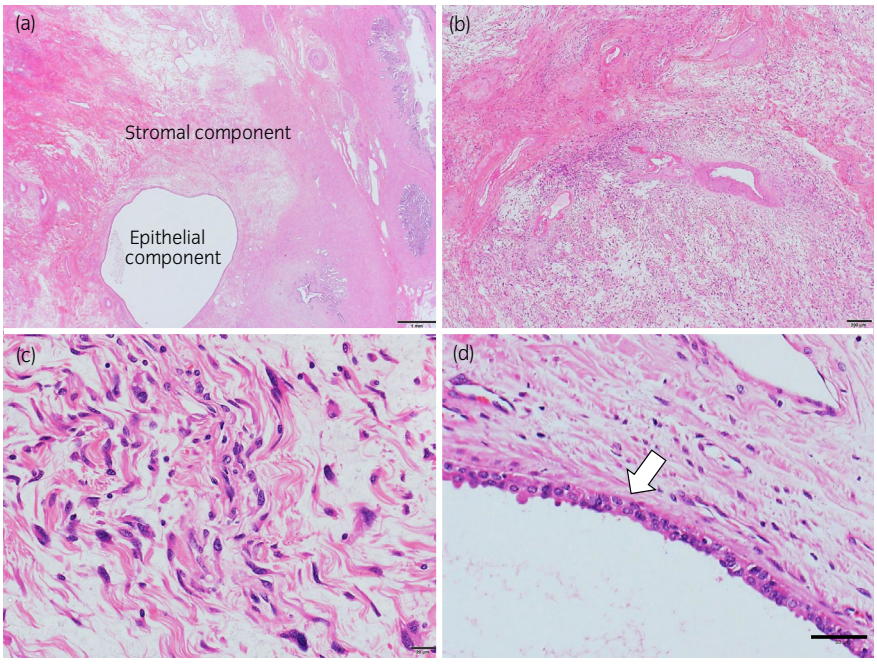

Fig. 2 Histological features of surgical specimen. (a) The tumor had stromal and epithelial components (×1.25, HE stain). (b) The stromal component was composed of spindle cells with varying degrees of cellularity (×4, HE stain). (c) Mild nuclear atypia and pleomorphism were focally present (×20, HE stain). (d) The epithelial component was composed of dilated, large, lined, cuboidal epithelial cells (×40, HE stain).

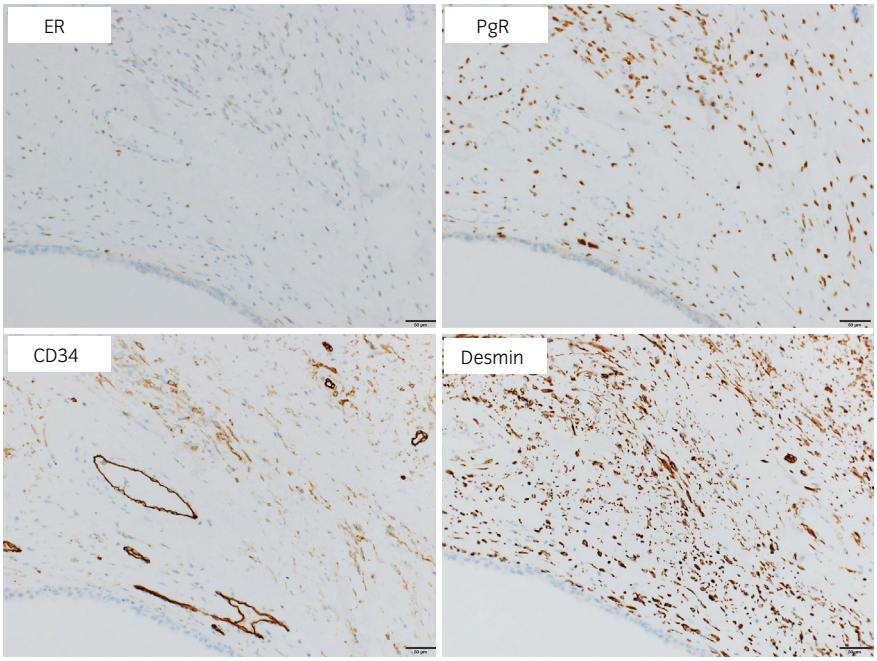

Fig. 3 Immunohistochemical findings of surgical specimen. Stromal component of the tumor was positive for estrogen receptor (weakly), progesterone receptor, CD34, and desmin.

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."[3] Pathologically, MEST is classified as low, intermediate, or high grade. Reikie *et al.*[1] 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,[4] 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.[5–7]

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,[8,9] and another two had lung metastases within 48 months, diagnosed as high grade.[10,11] One patient with high-grade disease died 11 months after metastatectomy.[10] 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.[12] Bullock also reported a 12-cm low-grade tumor with local recurrence 36 months after treatment due to incomplete resection.[9] 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.

Table 1 Summary of published cases of MEST from SV

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	Vesiculectomy	Low	60	NED
Lundhus *et al.*	1984	39	Cystadenoma	9	+ LUTS, abdominal/perineal pain	Vesicule-prostatectomy	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 (laparoscopic)	Low	36	Recurrence, 36 months after final resection
Raghuveer *et al.*	1989	45	Cystadenoma	5.5	+ 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	Vesiculectomy	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 MS, 6 months after chemotherapy
Peker *et al.*	1997	47	Cystadenoma	8	+ Hematospermia, suprapubic discomfort, tenesmus	Vesiculectomy	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	Vesiculectomy	High	11	Lung MS, died 11 months after resection
Gil *et al.*	2003	49	Cystadenoma	7	– S-DRE	Tumorectomy	Low	36	NED
Son *et al.*	2004	39	Phyllodes tumor	16	+ LUTS, abdominal pain	Vesiculectomy	Intermediate	24	NED
Lee *et al.*	2006	46	Cystadenoma	7.5	– S-DRE	Vesiculectomy	Low	6	NED
Hoshi *et al.*	2006	70	Epithelial stromal tumor	4.6	+ Abdominal pain, fatigue	Cystoprostatectomy/ileal neobladder	Low	14	NED
Khan *et al.*	2007	43	Phyllodes tumor	5.5	+ Hematospermia, testicular pain	Vesiculectomy (laparoscopic)	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	Vesiculectomy (laparoscopic)	Low	21	NED
Lorber *et al.*	2011	52	Cystadenoma	14	+ LUTS	Vesiculectomy (open)	Low	–	NA
Ploumidis *et al.*	2012	45	Cystadenoma	17.2	+ LUTS, pelvic pain	Vesiculectomy (robot-assisted)	Low	–	NA
Zhu *et al.*	2013	31	Cystadenoma	8.8	+ Hematospermia	Vesiculectomy (laparoscopic)	Low	–	NA
Arora *et al.*	2013	23	Cystadenoma	NA	+ LUTS, abdominal pain	Tumorectomy	Low	–	NA
Zhang *et al.*	2013	32	Cystadenoma	5	+ Hematospermia	Vesiculectomy (laparoscopic)	Low	19	NED
Zhang *et al.*	2013	64	Cystadenoma	4.5	+ Perineal pain	Vesiculectomy (laparoscopic)	Low	82	NED

Table 1 (Continued)

Author	Year	Age (years)	Author's terminology	Size (cm)		Symptom	Surgical approach	Grade	Follow-up (months)	Outcome
Zhang *et al.*	2013	50	Adenoma	3.8	+	Perineal pain	Vesiculectomy (laparoscopic)	Low	12	NED
Reikie *et al.*	2015	46	MEST	4	–	S-DRE	Vesicule-prostatectomy	Low	132	NED
Reikie *et al.*	2015	60	MEST	0.5	–	Radical prostatectomy	Vesicule-prostatectomy	Low	9	NED
Argun *et al.*	2015	48	Cystadenoma	6	+	Diminished ejaculate volume	Vesiculectomy (robot-assisted)	Low	12	NED
Campi *et al.*	2015	47	Cystadenoma	7	+	LUTS	Vesiculectomy (robot-assisted)	Low	24	NED
Kuai *et al.*	2017	71	Cystadenoma	6	–	Ultrasonography	Tumorectomy	Low	18	NED
Ameli *et al.*	2017	49	Cystadenoma	12	+	LUTS	Vesiculectomy (open)	Low	–	NA
Niu *et al.*	2017	59	Cystadenoma	7.5	+	LUTS	Vesiculectomy (laparoscopic)	Low	–	NA
Dong *et al.*	2018	37	Cystadenoma	11.9	+	Hematospermia, LUTS, hematuria	Vesiculectomy (laparoscopic)	Low	12	NED
Jaffer *et al.*	2018	32	Cystadenoma	14	+	LUTS	Vesiculectomy (open)	Low	6	NED
Tang *et al.*	2019	58	Cystadenoma	55	+	Hematospermia, LUTS, hematuria	Vesiculectomy (laparoscopic)	Low	–	NED
Current case	2019	52	MEST	3.2	–	CT	Vesicule-prostatectomy (robot-assisted)	Low	51	NED

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Conflict of interest

The authors declare no conflict of interest.

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