Submitted: 12 February, 2021 Accepted: 09 March, 2021 Published: 08 July, 2021 DOI:10.31083/jomh.2021.037

JOMH Journal of Men's Health

Case Report

Robot-assisted laparoscopic intracorporeal urachal mass resection and partial cystectomy for a huge urachal adenocarcinoma: a case report and review of literature

Jae Joon Park[1], Woong Bin Kim[2], Kwang Woo Lee[2], Jun Mo Kim[2], Young Ho Kim[2], Jae Heon Kim[1], Ahrim Moon[3], Si Hyun Kim[4], Sang Wook Lee[2,*]

[1]Department of Urology, Soonchunhyang University Seoul Hospital, Soonchunhyang University College of Medicine, 04401 Seoul, Korea
[2]Department of Urology, Soonchunhyang University Bucheon Hospital, Soonchunhyang University College of Medicine, 14584 Bucheon, Korea
[3]Department of Pathology, Soonchunhyang University Bucheon Hospital, Soonchunhyang University College of Medicine, 14584 Bucheon, Korea
[4]Department of Urology, Soonchunhyang University Cheonan Hospital, Soonchunhyang University College of Medicine, 31151 Cheonan, Korea

***Correspondence:** bartol@schmc.ac.kr (Sang Wook Lee)

Abstract

Urachal adenocarcinoma is rare, accounting for only 10% of adenocarcinomas of the bladder and the prognosis of urachal adenocarcinomas is poor since most cases are detected late. Since urachal adenocarcinoma is a rare disease, no effective standard treatment has yet been established. However, in recent studies, resection of carcinoma is considered the only treatment considered for non-metastatic cases. Although for large sized urachal adenocarcinoma, open surgery or laparoscopic surgery is usually considered, we have recently experienced huge urachal carcinoma by robotic surgery. We used cystoscopy and the robot to assess the cancer margins and safely perform the operation. A 71-year-old man with a medical history of hypertension and arrhythmia visited our urology department with urachal cancer detected by computed tomography (CT). CT showed a lobulated low-density mass, most likely urachal carcinoma, abutting the anterior dome of the bladder and anterior abdominal wall. We performed preoperative cystoscopy to assess the extent of the protrusion of the urachal cancer into the bladder wall and the area requiring resection during surgery. We confirmed the size and extent of the mass protruding into the anterior wall of the urinary bladder and Robot-assisted laparoscopic intracorporeal urachal mass resection and partial cystectomy using cystoscopy together was performed. After one month, the patient has no complications and no complaining symptoms complaints without any abnormal finding of follow up imaging test. Although more procedures must be performed to ensure the safety of robotic surgery as a treatment strategy for large urachal carcinomas, we confirm that robotic surgery can replace open or laparoscopic surgery for such tumors.

Keywords

Robot surgical procedure; Urachal adenocarcinoma; Minimal invasive surgical procedures; Case report

1. Introduction

The allantois degenerates during embryogenesis, and its vestigial structure form, the urachus, is a tubular structure that

connects the urinary bladder and umbilicus [1, 2]. During development, the urachus merges with the obliterated umbilical arteries to form the median urachal ligament and the remnants of the urachus degenerate. However, urachal remnants with epithelial-lined tubular or cystic muscle structures are found in approximately a third of adults. It is usually found in the dome of the urinary bladder and along the midline of the bladder wall [1, 3].

Bladder adenocarcinoma is rare, accounting for 0.5–2.0% of all bladder cancers [4]. However, urachal adenocarcinoma is even rarer, accounting for only 10% of adenocarcinomas of the bladder [1, 5]. Although urachal remnants and the bladder are composed of urothelial cells, there are several distinct differences between urachal and bladder carcinomas. Demographically, urachal carcinoma occurs in a younger patient population than that of bladder carcinoma, with a median diagnostic age of 47–56 years, and it is male dominant [3, 4, 6, 7]. Histologically, bladder carcinoma is a urothelial carcinoma, whereas most urachal carcinomas are adenocarcinomas expressed by a signet ring cell type [1, 8]. Among the histological forms of adenocarcinoma, the signet ring cell type related to urachal adenocarcinoma has the worst prognosis, with a 5-year overall survival rate of 27–61%. In addition, the prognosis of urachal carcinomas is poor since most are detected late, after the onset of hematuria [3].

Recently, we treated a patient with a huge urachal carcinoma by robotic surgery. We used cystoscopy and the robot to assess the cancer margins and safely perform the intracorporeal operation. This case report was conducted in accordance with the Declaration of Helsinki. Informed written consent was obtained from the patient for publication of this report and any accompanying images. The authors have read the CARE Checklist, and the manuscript was prepared and revised according to the CARE Checklist.

2. Case presentation

2.1 Patient information and clinical findings

A 71-year-old man with a medical history of hypertension and arrhythmia visited our urology department with urachal cancer detected by computed tomography (CT), incidentally. The patient had inadvertently felt a lump in his lower abdomen and visited a local hospital. CT performed at the local hospital showed a lobulated low-density mass, most likely urachal carcinoma, abutting the anterior dome of the bladder and anterior abdominal wall (Fig. 1). The patient visited our urology department for evaluation of these abnormal findings. On physical examination, a large non-tender mass was palpable in the patient's lower abdomen. No microscopic hematuria was observed in urine analysis.

2.2 Diagnostic assessment

We performed preoperative cystoscopy to assess the extent of the protrusion of the urachal cancer into the bladder wall and the area requiring resection during surgery. We confirmed the size and extent of the mass protruding into the anterior wall of the urinary bladder (Fig. 2). Considering our recommendations, the patient decided to undergo radical resection of the urachal cancer and partial cystectomy.

2.3 Surgical procedure

The surgeon was experienced in robot-assisted laparoscopic radical prostatectomy and partial nephrectomy. The da Vinci® Xi system (Intuitive Surgical, Inc., Sunnyvale, CA, USA) was used for the operation, which was performed under general anesthesia. A nasogastric tube was placed before positioning. The patient was placed in the lithotomy and Trendelenburg positions. The abdomen and pelvic area were prepped and draped in a conventional sterile manner. Cystoscopy was performed to recheck the mass margins in the bladder, and the margins were marked using bipolar electrode. A robotic optical trocar was inserted into the abdomen through a camera port via a 1 cm supraumbilical incision. The robotic trocar and assistant port were arranged horizontally with the umbilicus at 8 cm intervals. After robot docking, the mass (Fig. 3A) was visually identified and separated from the abdominal wall using electrocautery. Cystoscopy was performed again after excising all of the mass except the part connected to the bladder. The mass margins were checked using the cystoscopic and robotic visual fields together, and the remaining mass protruding into the bladder was resected. During this process, we could see the cystoscopic light transmitted through the bladder wall in the robotic visual field, which was helpful in accurately identifying the mass margins and securing a cancer-negative margin (Fig. 3B). In addition, the demarcation line placed in the bladder was helpful in checking and resecting the margins (Fig. 3C). The bladder was reconstructed in a double layer (Fig. 3D) and filled with 200 mL saline to confirm that there was no leakage. A Jackson-Pratt drain (Cardinal Health, Waukegan, IL, USA) was positioned and anchored, and the skin wound was closed layer by layer.

2.4 Pathologic findings

The entire specimen was 13.0 × 9.0 × 4.5 cm in size and contained an 8.0 × 8.0 × 5.0 cm mucinous mass (Fig. 4A). The tumor extended to the bladder and subserosa, with a negative resection margin. Well-differentiated mucin-producing adenocarcinoma cells were seen on hematoxylin and eosin (H&E) stained specimens at 20× magnification (Fig. 4B). Tumor cells floating in mucin lakes were seen on H&E stained specimens at 100× magnification (Fig. 4C). Immunohistochemically, the tumor tissue was positive for CDX2 (Fig. 4D). The final histopathological diagnosis was urachal adenocarcinoma.

2.5 Outcome and follow-up

The surgical duration was 150 mins, and the estimated blood loss was 50 mL. The Jackson-Pratt drain was removed on postoperative day 3, and all stitches were removed on postoperative day 12. On postoperative day 13, we performed cystography, which confirmed no abnormal findings such as urine leakage (Fig. 5). The Foley catheter was removed, and

JOMH
Journal of Men's Health

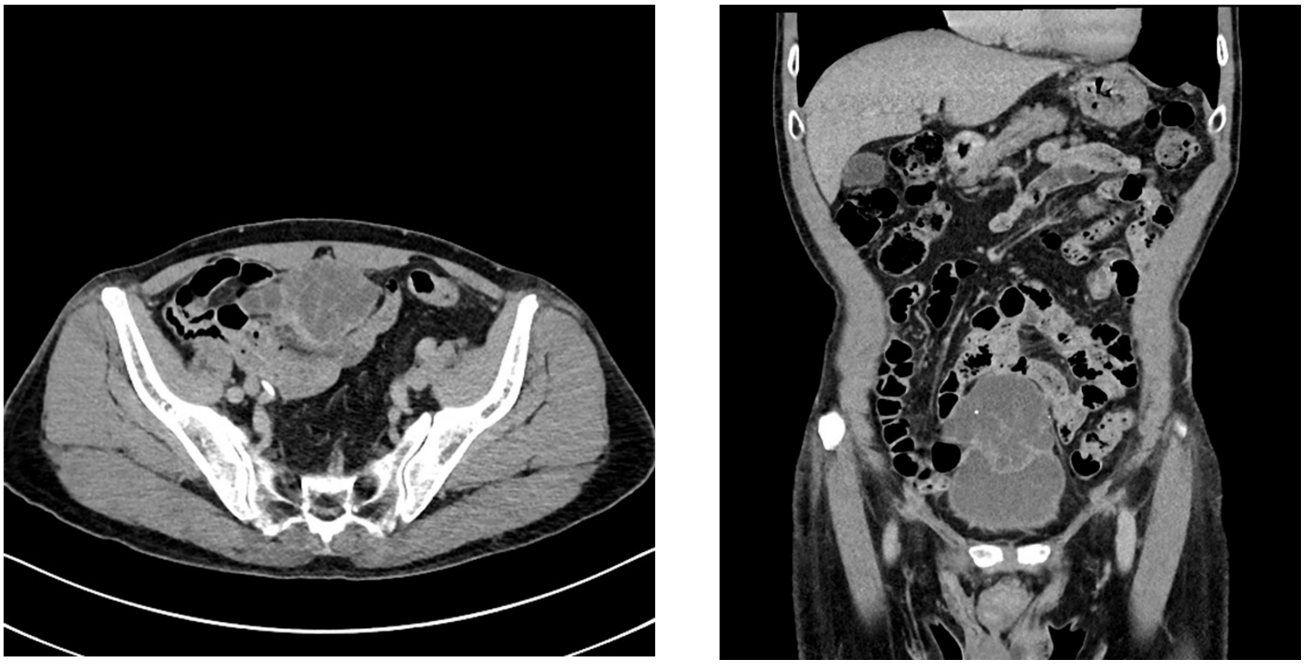

FIG. 1. Preoperative computed tomography showed a lobulated low-density mass, most likely urachal carcinoma, abutting the anterior dome of the bladder and anterior abdominal wall.

FIG. 2. The mass protruding into the anterior wall of the bladder was confirmed by preoperative cystoscopy.

the patient was discharged on postoperative day 14 with no complications.

On postoperative day 28, follow-up CT showed no specific findings other than irregular wall thickening of the anterior and dome areas of the urinary bladder as a postoperative change (Fig. 6). There were no complications or complaints of symptoms by the patient for 3 months. Further follow-up CT was planned after 3 months.

3. Discussion

Primary urachal adenocarcinoma is a rare type of carcinoma first reported by Hue and Jacquin in 1863. Urachal remnants, found in approximately one-third of adults, are composed of three layers: an outer smooth muscle layer, intermediate submucosal connective tissue layer, and inner luminal layer. The cells of these three layers, particularly epithelial cells, cause urachal carcinoma [3, 7, 9]. Most urachal carcinomas are adenocarcinomas; the reason for this is not clear, but glandular metaplasia in urachal urothelium may be the cause of adenocarcinomas [1, 7].

The diagnosis of urachal carcinoma should be considered after taking a thorough medical history and performing a physical examination [10]. Hematuria is the most common symptom, generally suggesting an advanced stage, and is a predictor of malignancy and bladder involvement [3, 10]. The extents of the mass and local lymph node metastasis are assessed using abdominal pelvic CT or magnetic resonance imaging, and metastases are further evaluated by chest radiographs or bone scans. Urine cytology can also be helpful for diagnosis, and cystoscopy is necessary for determining if the carcinoma has penetrated the bladder urothelium, and the need for transurethral biopsy [10].

In addition, histology is also helpful. According to Grignon *et al.* [11], urachal carcinomas are classified into the following five histological subtypes: intestinal, mucinous, signet ring cell, unspecified, and mixed. It is essential to differentiate between urachal and non-urachal adenocarcinomas of the bladder because of differences in the treatment methods between them. However, since urachal and non-urachal adenocarcinomas are both positive for CK7, CK20, and CDX2, immunohistochemical approaches alone are limited. Therefore, a comprehensive evaluation combining pathology, radiological imaging, and cystoscopic findings is necessary [1, 12].

There have been several changes in the diagnostic criteria for urachal tumors. Initially, Wheeler and Hill established rigorous standards that required multiple criteria to be met, such as presence of a tumor in the dome of the bladder, absence of cystitis cystica, presence of urachal remnants, and presence of a suprapubic mass [1, 13]. However, because these strict criteria run the risk of excluding many reported

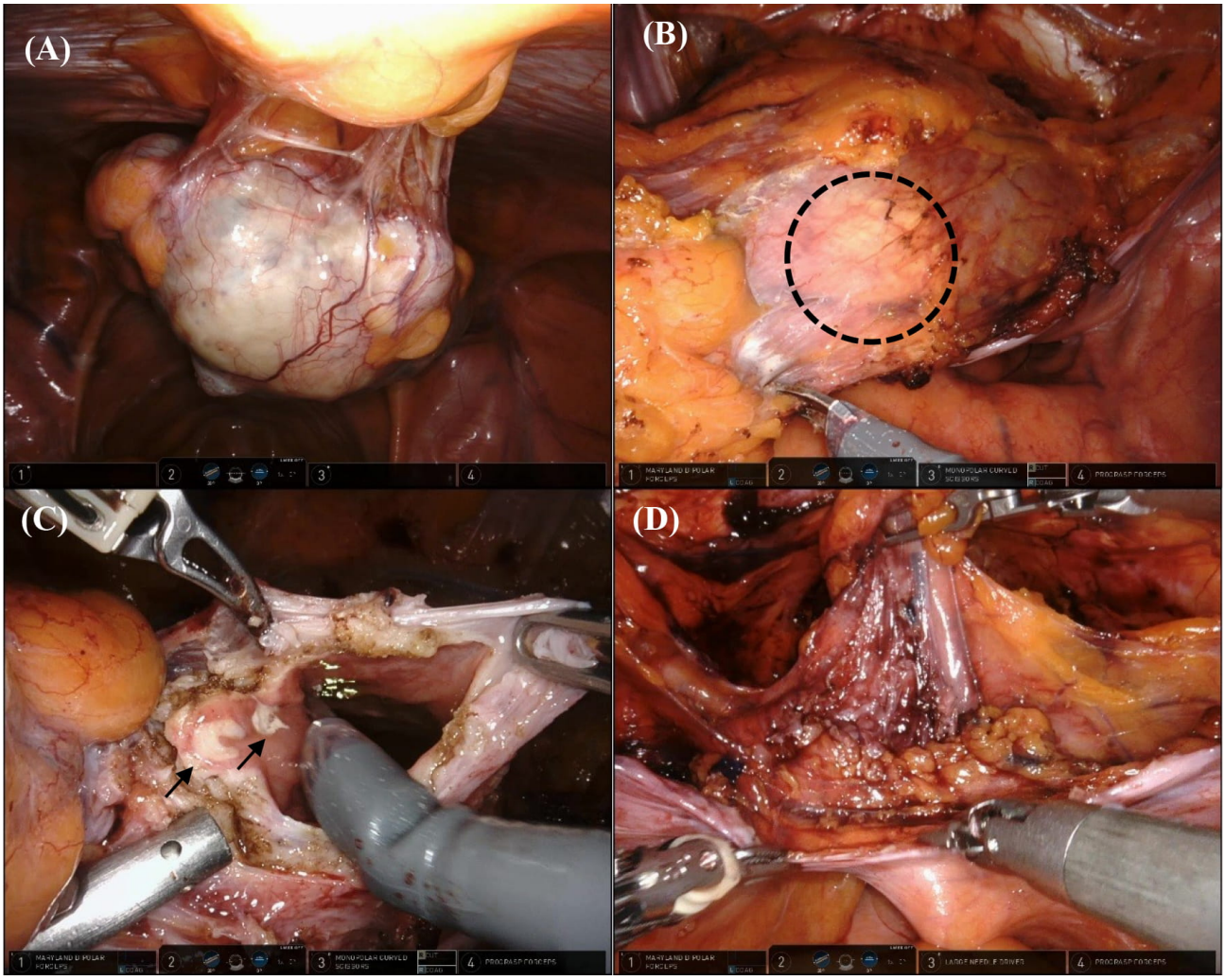

FIG. 3. Intraoperative gross findings. A huge urachal mass was observed in the abdominal cavity (A). Using the cystoscope light (dotted circle) transmitted through the bladder wall, excision of the mass was performed (B). The mass was resected along the demarcation line (arrows) in the bladder (C). After resection of the mass, the bladder was reconstructed in a double layer (D).

urachal carcinomas, Johnson *et al.* [14] modified the criteria for urachal carcinomas; presence of a tumor in the dome or elsewhere in the midline of the bladder, sharp demarcation between the tumor and surface epithelium, and absence of a primary adenocarcinoma of another organ [1].

There are several staging systems for urachal carcinoma [15, 16]. Sheldon *et al.* [15] proposed a staging system in which stages I and II are defined by the presence of a urachal remnant. However, because the number of patients with urachal remnants is small, and the number of cystectomy specimens containing urachal remnants is as low as 17.5%, most cases were judged to be above stage III. Another staging system proposed by Ashley *et al.* [16] of the Mayo Clinic had trouble distinguishing between stages III and IV. The TNM staging system for bladder carcinomas was initially thought to be limited for the evaluation of urachal carcinoma because this tumor does not occur in the bladder urothelium. However, studies have shown that this system may be useful for evaluating urachal carcinoma in terms of tumor extent and metastasis to the bladder wall [1].

Since urachal adenocarcinoma is a rare disease, no effective standard treatment has yet been established [3, 10, 17]. A retrospective study by Pinthus *et al.* [18] confirmed that surgical treatment results in a higher survival rate than non-surgical treatment. Among surgical treatments, a study by Bruin *et al.* [19] comparing radical and partial cystectomy showed no significant difference in survival rate, and therefore extensive resection is the only treatment considered for non-metastatic cases. It is necessary to ensure a cancer-negative surgical margin using partial cystectomy with en bloc resection of the urachus and surrounding peritoneum, and some studies recommend additionally performing umbilectomy in all cases [16, 17, 20]. The effectiveness of adjuvant chemotherapy or adjuvant radiotherapy for urachal carcinoma is not clear. A study by Ashley *et al.* [16] confirmed that chemotherapy and radiotherapy had no significant effect on the survival rate of patients with urachal carcinoma [20]. Minimally invasive surgical methods such as laparoscopic or robotic surgery are also considered options for urachal carcinoma but have rarely been reported. For this approach, oncological knowledge and sufficient robotic surgical expertise are required [21, 22].

Similar cases have been reported before. Kosanovic *et al.*

JOMH
Journal of Men's Health

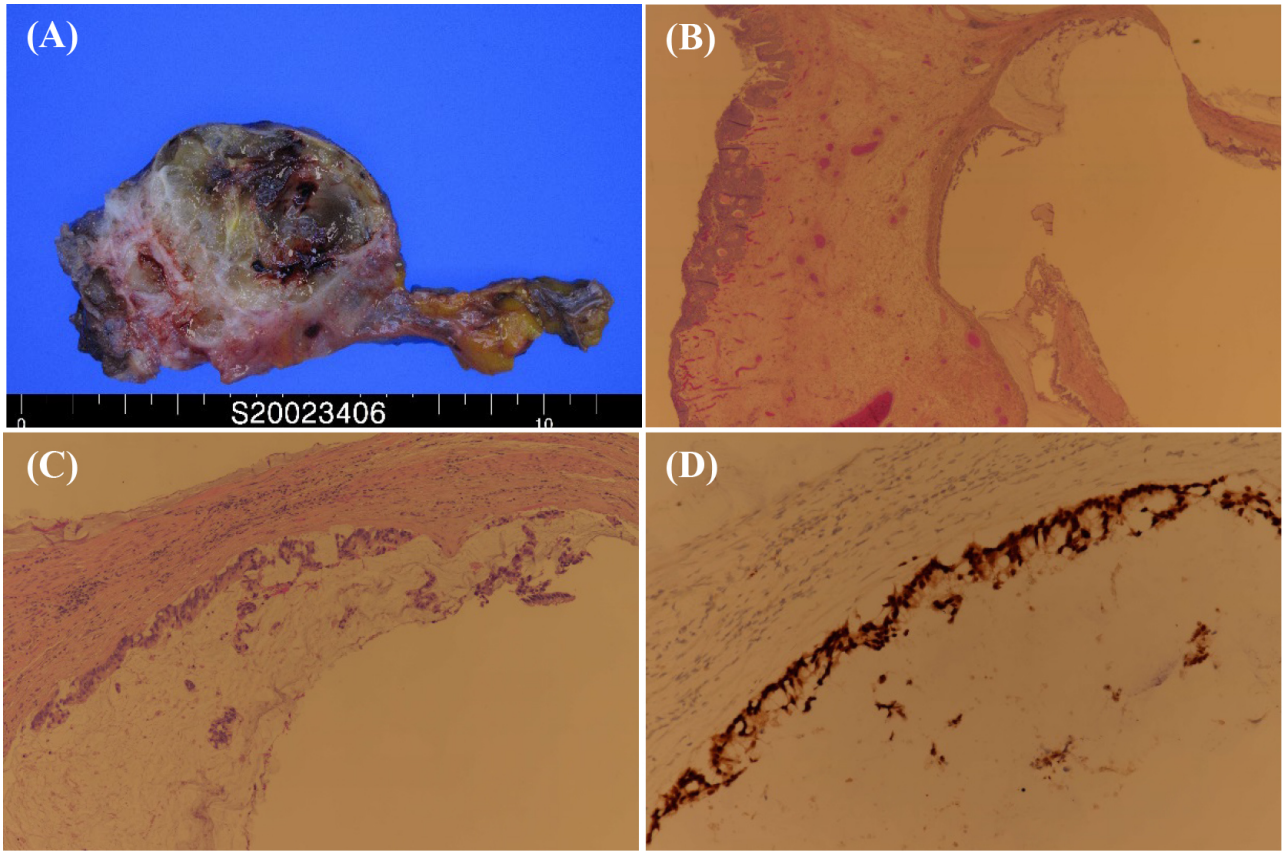

FIG. 4. Pathologic findings. An 8.0 × 8.0 × 5.0 cm mucinous mass was detected within the resected tumor (A). Well-differentiated mucin-producing adenocarcinoma cells were seen in hematoxylin and eosin (H&E) stained specimens at 20× magnification (B). Tumor cells floating in mucin lakes were present in H&E stained specimens at 100× magnification (C). Immunohistochemically, the tumor was positive for CDX2 (D).

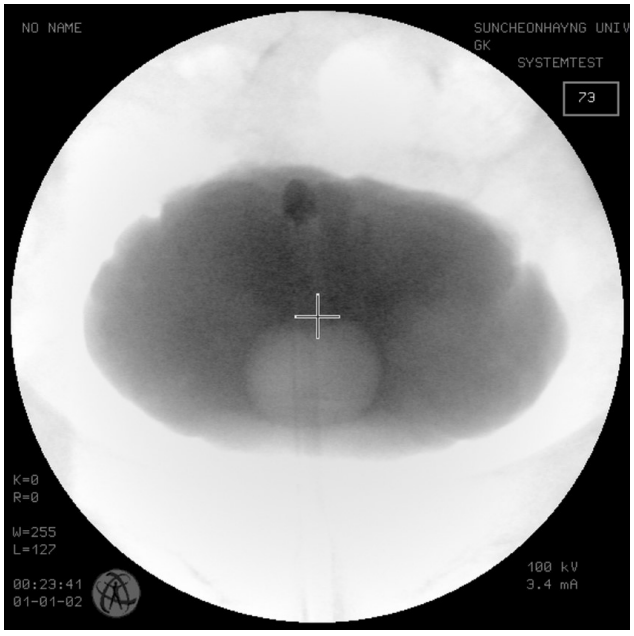

FIG. 5. Cystography on postoperative day 14 showed no abnormal findings.

performed robotic resection of a urachal tumor on urachal mucinous adenocarcinoma of 5.4 × 6.7 × 5.9 cm in 2014. Later, as a result of pathology examination, since the viable tumor was close to the margin of resection, resection of the umbilicus, urachal remnant and bladder dome was performed 4 months later [23]. Aoun *et al.* performed a bladder sparing robot-assisted laparoscopic en bloc resection of urachus on 23 × 21 × 26 mm urachal carcinoma in 2015, and also used flexible cystoscopy to confirm the endoluminary boundary of the mass. It was maintained without recurrence at follow up after 3 months [24]. Quan *et al.* reported two cases of urachal carcinoma in 2017. A radical resection of the urachal tumor and partial cystectomy was performed, and no recurrence was found at follow up after 10 months [3]. As such, several similar cases have been reported, but there are several noticeable points only in this case. First, the size of urachal adenocarcinoma in this case is 8.0 × 8.0 × 5.0 cm, which is the largest among robotic surgery cases reported. Introducing the process and results of robotic tumor resection performed on such large-sized tumors is thought to be helpful in determining treatment methods in other urachal carcinoma cases. Second, participation by a co-author, who is pathologist, presented a more detailed pathological analysis.

In the present case, the patient has experienced no complications during follow-up to date. Since the bladder is located deep in the pelvic cavity and has a complex anatomical relationship with surrounding organs, it is difficult to secure adequate vision. Robotic surgery can reduce the risk of surgical site infections risk and can minimize damage to peripheral organs, blood vessels, and nerves because it has the advantage

FIG. 6. Computed tomography on postoperative day 28 showed no specific findings other than irregular wall thickening of the anterior and dome areas of the urinary bladder as a postoperative change.

of achieving sufficient field of vision [25]. The robot based minimally invasive approach, which has proven its safety and efficacy in the treatment of other tumors in the urology field, is already widely being used [26]. The intracorporeal robotic surgery for urachal carcinoma also has the advantages of minimizing the operative incision, relatively little bleeding, easy bladder closure, and a quick return to daily life for the patient. In addition, if cystoscopy is used for robotic surgery as in this case, the tumor margin can be directly assessed and accurately excised, and safe surgery is possible even with very large masses. Despite these advantages, the limitation of this case report was short follow-up period. A further well-designed study is needed to overcome this limitation.

4. Conclusions

Although more procedures must be performed to ensure the safety of robotic surgery as a treatment strategy for large urachal carcinomas, we confirm that robotic surgery can replace open or laparoscopic surgery for such tumors. This procedure can safely and accurately remove the mass, reduce postoperative complications based on a superior field of view, and secure the mass margins by simultaneous use of cystoscopy. In this case, a huge urachal adenocarcinoma was safely excised.

Abbreviations

CT, computed tomography; H&E, hematoxylin and eosin.

Author contributions

Conceptualization: JJP, SWL. Data curation: JJP, SHK, WBK. Methodology: KWL, JMK, YHK, SWL. Validation: SHK, JHK. Investigation: JJP, AM, SWL. Writing—original draft: JJP, SWL. Writing—review & editing: SHK, SWL.

Ethics approval and consent to participate

This case report was conducted in accordance with the Declaration of Helsinki. Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Acknowledgment

1. This manuscript was prepared with the assistance of TEXTCHECK, an English-language scientific editing company.
2. This manuscript titled "Robot-assisted laparoscopic intracorporeal urachal mass resection and partial cystectomy for a huge urachal adenocarcinoma: A case report and review of literature" has been edited to ensure that the language is clear and free of errors. The edit was performed by Dr. Wilson Kim.
3. Thanks to all the peer reviewers for their opinions and suggestions.

Funding

This work was supported by the "Soonchunhyang University Research Fund".

Conflict of interest

The authors declare no conflict of interest.

References

[1] Dhillon J, Liang Y, Kamat AM, Siefker-Radtke A, Dinney CP, Czerniak B, *et al.* Urachal carcinoma: a pathologic and clinical study of 46 cases. Human pathology. 2015; 46: 1808–1814.
[2] Begg RC. The urachus: its anatomy, histology and development. Journal of Anatomy. 1930; 64: 170.
[3] Quan J, Pan X, Jin L, He T, Hu J, Shi B, *et al.* Urachal carcinoma: report of two cases and review of the literature. Molecular and clinical oncology. 2017; 6: 101–104.

[4] Wright JL, Porter MP, Li CI, Lange PH, Lin DW. Differences in survival among patients with urachal and nonurachal adenocarcinomas of the bladder. Cancer. 2006; 107: 721–728.
[5] Herr HW, Bochner BH, Sharp D, Dalbagni G, Reuter VE. Urachal carcinoma: contemporary surgical outcomes. The Journal of urology. 2007; 178: 74–78.
[6] Siefker-Radtke A. Urachal adenocarcinoma: a clinician's guide for treatment. *In* Seminars in oncology (pp. 619–624). Elsevier. 2012.
[7] Gopalan A, Sharp DS, Fine SW, Tickoo SK, Herr HW, Reuter VE, *et al.* Urachal carcinoma: a clinicopathologic analysis of 24 cases with outcome correlation. The American journal of surgical pathology. 2009; 33: 659.
[8] Dinney CP, McConkey DJ, Millikan RE, Wu X, Bar-Eli M, Adam L, *et al.* Focus on bladder cancer. Cancer cell. 2004; 6: 111–116.
[9] Nix J, Menville J, Albert M, Wendt D. Congenital patent urachus. The Journal of urology. 1958; 79: 264–273.
[10] Williams CR, Chavda K. En bloc robot-assisted laparoscopic partial cystectomy, urachal resection, and pelvic lymphadenectomy for urachal adenocarcinoma. Reviews in urology. 2015; 17: 46.
[11] Grignon DJ, Ro JY, Ayala AG, Johnson DE, Ordóñez NG. Primary adenocarcinoma of the urinary bladder. A clinicopathologic analysis of 72 cases. Cancer. 1991; 67: 2165–2172.
[12] Paner GP, McKenney JK, Barkan GA, Yao JL, Frankel WL, Sebo TJ, *et al.* Immunohistochemical analysis in a morphologic spectrum of urachal epithelial neoplasms: diagnostic implications and pitfalls. The American journal of surgical pathology. 2011; 35: 787–798.
[13] Wheeler J, Hill WT. Adenocarcinoma involving the urinary bladder. Cancer. 1954; 7: 119–135.
[14] Johnson DE, Hodge GB, Abdul-Karim FW, Ayala AG. Urachal carcinoma. Urology. 1985; 26: 218–221.
[15] Sheldon CA, Clayman RV, Gonzalez R, Williams RD, Fraley EE. Malignant urachal lesions. The Journal of urology. 1984; 131: 1–8.
[16] Ashley RA, Inman BA, Sebo TJ, Leibovich BC, Blute ML, Kwon ED, *et al.* Urachal carcinoma: clinicopathologic features and long-term outcomes of an aggressive malignancy. Cancer. 2006; 107: 712–720.
[17] Siefker-Radtke A. Urachal carcinoma: surgical and chemotherapeutic options. Expert review of anticancer therapy. 2006; 6: 1715–1721.
[18] Pinthus JH, Haddad R, Trachtenberg J, Holowaty E, Bowler J, Herzenberg AM, *et al.* Population based survival data on urachal tumors. The Journal of urology. 2006; 175: 2042–2047.
[19] Bruins HM, Visser O, Ploeg M, Hulsbergen-van de Kaa CA, Kiemeney LA, Witjes JA. The clinical epidemiology of urachal carcinoma: results of a large, population based study. The Journal of urology. 2012; 188: 1102–1107.
[20] Asano K, Miki J, Yamada H, Maeda S, Abe K, Furuta A, *et al.* Carcinoma of urachus: report of 15 cases and review of literature–is total cystectomy the treatment of choice for urachal carcinoma? Nihon Hinyokika Gakkai zasshi. The japanese journal of urology. 2003; 94: 487.
[21] Spiess PE, Correa JJ. Robotic assisted laparoscopic partial cystectomy and urachal resection for urachal adenocarcinoma. International Brazilian Journal of Urology. 2009; 35: 609.
[22] Madeb R, Knopf JK, Nicholson C, Donahue LA, Adcock B, Dever D, *et al.* The use of robotically assisted surgery for treating urachal anomalies. BJU international. 2006; 98: 838–842.
[23] Kosanovic R, Romero RJ, Arad JK, Gallas M, Seetharamaiah R, Gonzalez AM. Rare use of robotic surgery for removal of large urachal carcinoma. Journal of robotic surgery. 2014; 8: 177–180.
[24] Aoun F, Peltier A, Van Velthoven R. Bladder sparing robot-assisted laparoscopic en bloc resection of urachus and umbilicus for urachal adenocarcinoma. Journal of robotic surgery. 2015; 9: 167–170.
[25] de Vermandois JAR, Cochetti G, Del Zingaro M, Santoro A, Panciarola M, Boni A, *et al.* Evaluation of surgical site infection in mini-invasive urological surgery. Open Medicine. 2019; 14: 711–718.
[26] Mearini E, Cirocchi R, Cochetti G. Robot-assisted surgery in urology: the show must go on. Multidisciplinary Digital Publishing Institute. 2019.