


## Case Report

# Combination of en bloc transurethral resection with laparoscopic partial cystectomy for paraganglioma of the bladder

Fumihiko Urabe,<sup>1,2</sup>  Jun Miki,<sup>1</sup> Takahiro Kimura,<sup>2</sup> Akira Hisakane,<sup>1</sup> Kosuke Iwatani,<sup>1</sup> Kazuhiro Takahashi,<sup>1</sup> Takafumi Yanagisawa,<sup>1</sup> Hiroyuki Takahashi,<sup>3</sup> Koichi Kishimoto<sup>1</sup> and Shin Egawa<sup>2</sup>

<sup>1</sup>Department of Urology, The Jikei University Kashiwa Hospital, Kashiwa, Chiba, Departments of <sup>2</sup>Urology, and <sup>3</sup>Pathology, The Jikei University School of Medicine, Minato-ku, Tokyo, Japan

### Abbreviations & Acronyms

CT = computed tomography  
LECS = laparoscopic and endoscopic cooperative surgery  
MIBG = metaiodobenzylguanidine  
MRI = magnetic resonance imaging  
TUR = transurethral resection  
TUR-BT = transurethral resection of bladder tumor

**Correspondence:** Fumihiko Urabe M.D., Department of Urology, The Jikei University School of Medicine, 3-25-8 Nishi-Shimbashi, Minato-ku, Tokyo 105-8471, Japan. Email: furabe0809@gmail.com

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**Introduction:** Paraganglioma of the bladder is an extremely rare tumor, and special attention should be paid to elevation of blood pressure during tumor resection.

**Case presentation:** A 64-year-old woman presented with elevation of blood pressure during thoracic surgery. Multiparametric magnetic resonance imaging revealed a bladder tumor, and noradrenalin levels were elevated in plasma and urine. The tumor was surgically removed by en bloc transurethral resection with laparoscopic partial cystectomy of bladder tumor. The margin of the tumor was easily identified, and perioperative blood pressure was stable. There was no evidence of residual tumor, local recurrence, or distant metastasis during 2-year follow-up.

**Conclusion:** To our knowledge, we report the first case of paraganglioma of the bladder resected by a minimally invasive and safe procedure: combination of en bloc transurethral resection and laparoscopic partial cystectomy.

**Key words:** en bloc TUR, laparoscopic surgery, LECS, paraganglioma, pheochromocytoma.

## Keynote message

We report a minimally invasive and safety approach for the resection of bladder paraganglioma.

## Introduction

Pheochromocytoma is a chromaffin cell tumor usually arising in the adrenal glands and is characterized by excessive production of catecholamines, often leading to elevation of blood pressure. Only <15% of pheochromocytomas develop from extra-adrenal chromaffin tissues, which are classified as paraganglioma. Paraganglioma of the bladder is extremely rare and originates from the chromaffin tissues of the sympathetic nervous system in the bladder wall.<sup>1</sup> During tumor resection, special attention should be paid to the elevation of blood pressure. Here, we report a minimally invasive and safe procedure to resect paraganglioma of the bladder using endoscopic and laparoscopic cooperative approach: combination of en bloc TUR and laparoscopic partial cystectomy.

## Case presentation

A 64-year-old woman had undergone video-assisted lobectomy for lung cancer and developed elevation of systolic blood pressure up to 300 mmHg. Although an adrenal mass was not detected, thoracoabdominal CT revealed a 2.0 × 2.1-cm soft tissue mass protruding from the posterior aspect of the bladder (Fig. 1a). Multiparametric MRI of the abdomen and pelvis demonstrated a 2.0 × 2.1-cm soft tissue mass in the same place. The lesion had a high T1-weighted intensity and low T2-weighted signal intensity (Fig. 1b). Hormone analysis revealed elevation of plasma noradrenaline (670 pg/mL), urinary noradrenaline (321 g/day), urinary

dopamine (2100 µg/day), and urinary metanephrine (0.38 mg/day) levels. 131I-MIBG scintigraphy revealed uptake of 131I-MIBG by the tumor (Fig. 1c). No additional lesions to support metastases were found on performing CT scan or 131I-MIBG scintigraphy. Cystoscopy revealed a solitary submucosal mass on the posterior aspect of the bladder, with a grossly normal mucosa (Fig. 1d). From these results, a diagnosis of paraganglioma of the bladder was made, and we decided to perform surgical resection of the tumor.

Under general anesthesia, we first performed en bloc TUR. Briefly, we made circular incisions around the tumor using a needle electrode under a transurethral scope (Fig. 2a). Careful dissection of bladder muscle layers was performed using pull incision not to rupture the bladder (Fig. 2b,c). Next, using a three-port (5–10 mm) triangular arrangement at a 15° Trendelenburg position, we performed transperitoneal laparoscopic partial cystectomy. We cut the peritoneum and bladder serosa (Figs 2d,3a) and removed the specimen using a laparoscopic sac (Fig. 3b). We confirmed that both ureters were intact and sutured the bladder wall using a 3-0 monofilament polyglyconate suture (Fig. 3c). Distension of the bladder at the completion of the repair showed no leakage (Fig. 3d). A drain was placed in the pelvis, and a 16-Fr Foley catheter was indwelled into the bladder through the urethra. During the operation, blood pressure was stable (systolic blood pressure range: 80–130 mmHg without drastic fluctuation). The total surgical time was 137 min. The drain was removed 5 days after surgery.

The tumor extracted was 1.8 × 1.6 × 1.2 cm in size (Fig. 4a). Pathological analysis revealed polygonal tumor cells with fine granular cytoplasm present in the submucosa and muscularis propria on performing hematoxylin and eosin staining (Fig. 4b,c). These findings were consistent with the

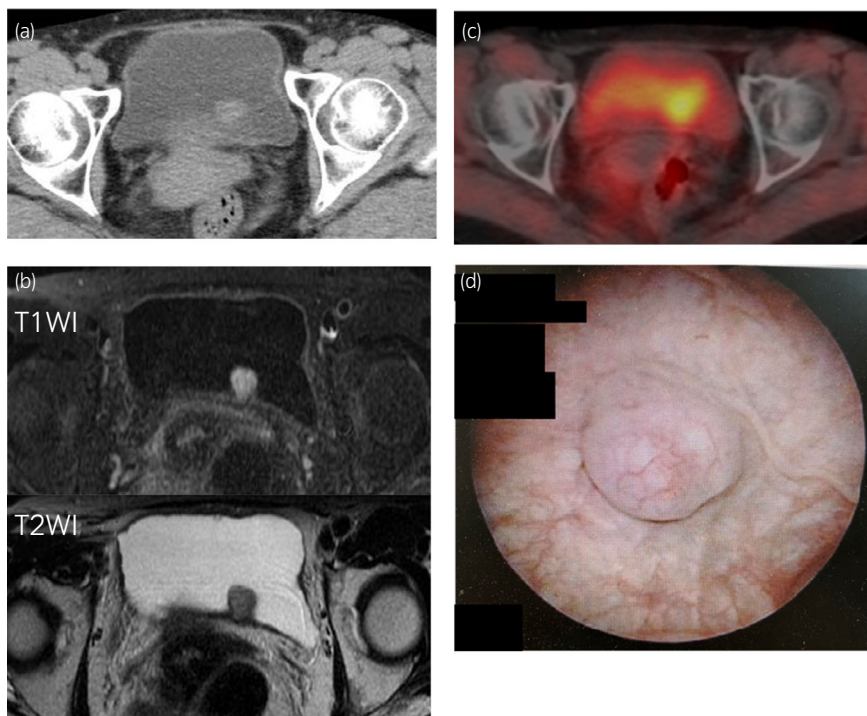
diagnosis of paraganglioma of the bladder. Lymphovascular invasion was not observed. Surgical margins were free of tumor.

One month after surgery, her endocrine panel was normal. There was no evidence of residual tumor, local recurrence, or distant metastasis during 2-year follow-up.

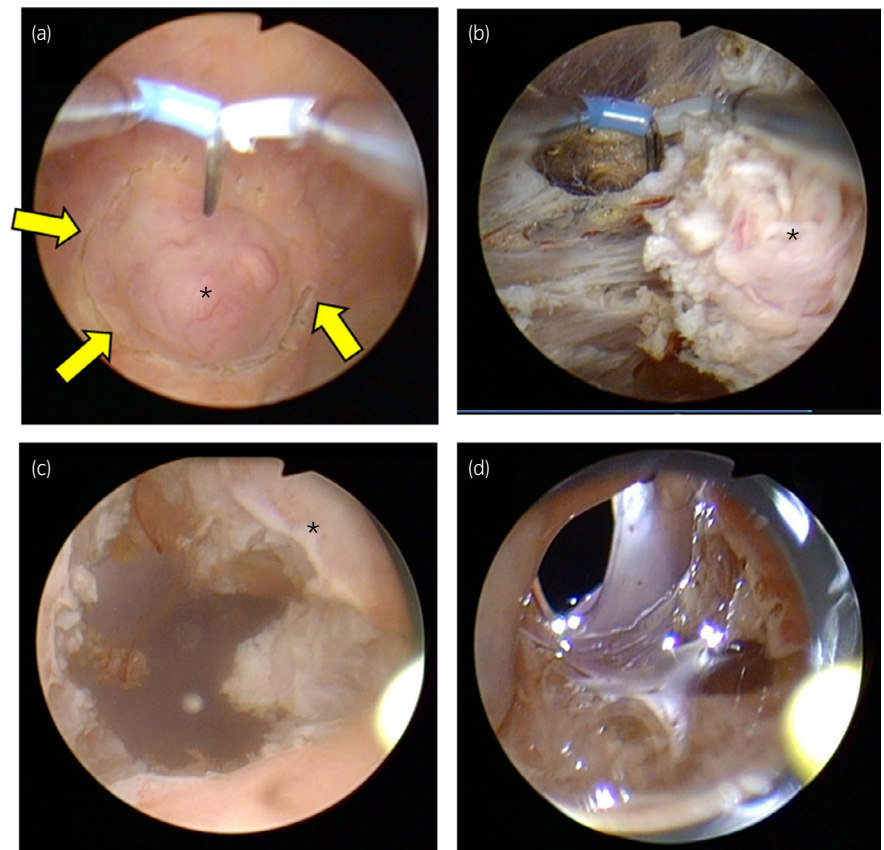
## Discussion

Paraganglioma of the bladder is extremely rare and accounts for about 0.05% of all bladder tumors and <1% of all pheochromocytomas.<sup>2,3</sup> Paraganglioma of the bladder is a neuroendocrine tumor and typically arises from the chromaffin tissue of the sympathetic nervous system in the muscular layer of the bladder wall.<sup>4</sup> Although in this case, the findings of MRI were atypical; high intensity in T1-weighted and low intensity in T2-weighted, we could diagnose as paraganglioma of the bladder from MIBG and hormone examination. As the appearance of MRI images of paraganglioma can be varied, paraganglioma is recommended not to be excluded on the basis of a lack of high signal intensity at T2-weighted MR imaging.<sup>5</sup>

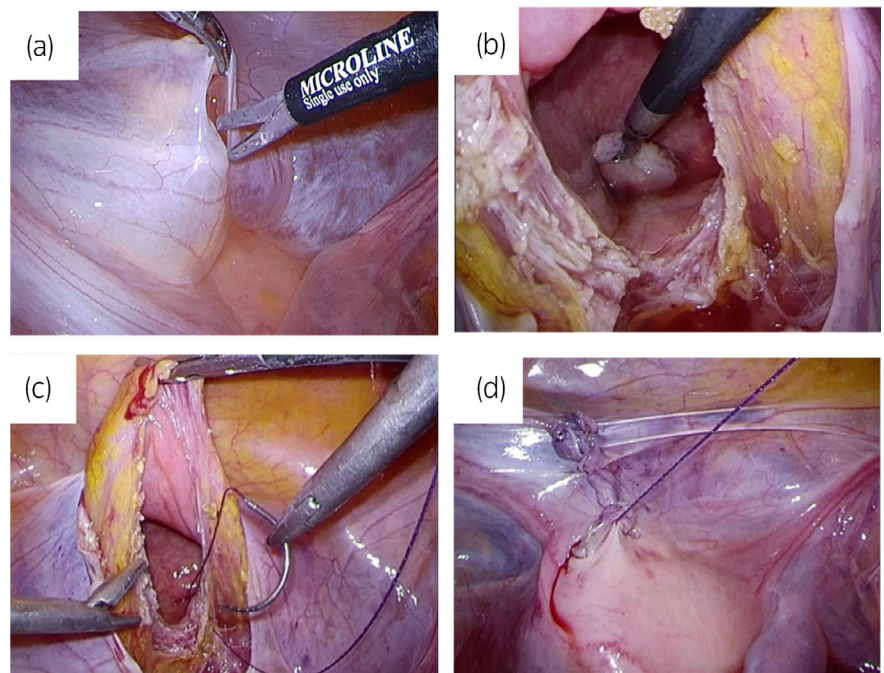
Treatment of paraganglioma of the bladder may include various methods, such as catecholamine blockade, surgery, radiation therapy, and chemotherapy. However, surgical resection remains the standard treatment.<sup>6</sup> TUR-BT and cystectomy (partial or radical) were the main methods used for the resection of tumor. However, TUR-BT can cause elevation of blood pressure during surgery. Iwamoto *et al.* reported sharp fluctuations in blood pressure at the start of TUR for paraganglioma of the bladder.<sup>7</sup> Recently, laparoscopic or robot-assisted laparoscopic partial cystectomy has become the most commonly reported treatment for patients



**Fig. 1** (a) CT scanning showing 2.0 cm soft tissue mass protruding from the posterior bladder wall, (b) the lesion had a high T1-weighted image and low T2 image signal intensity on MRI, (c) 131I-MIBG scintigraphy showing uptake of 131I-MIBG by the tumor, (d) cystoscopy revealed a solitary submucosal mass with normal mucosal covering.



**Fig. 2** Cystoscopic findings of the en bloc TUR. (a) Circular incision around the tumor using needle electrode, (b) dissection of bladder muscle layer using pull incision, (c) after completion of dissection, (d) cystoscopic view after transperitoneal laparoscopic incision of peritoneum and bladder serosal layer.

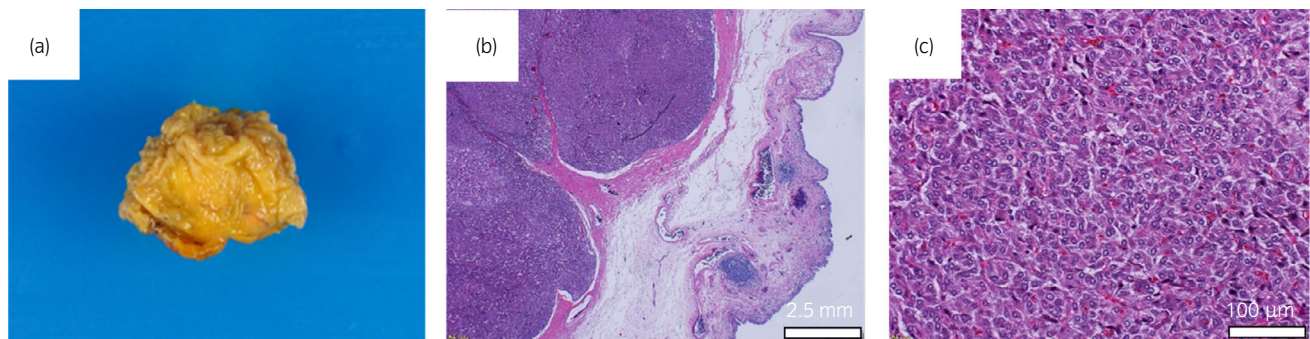


**Fig. 3** (a) Transperitoneal laparoscopic incision of peritoneum and bladder serosal layer, (b) laparoscopic view of partial cystectomy, (c) bladder repair using a 3-0 monofilament polyglyconate suturing, (d) leakage test.

with paraganglioma of the bladder.<sup>6,8,9</sup> In comparison with open surgery, laparoscopic surgery offers the following advantages: minimally invasive, faster recovery, and shorter hospitalization.<sup>10</sup> However, clearly detecting the tumor margin through intraperitoneal laparoscope remains a challenge.

In the present case, we performed the resection of the paraganglioma by combination of en bloc TUR and laparoscopic partial cystectomy. Compared to the conventional methods, the operators could easily identify the resection margin of the tumor under cystoscopy and transperitoneal laparoscopy,





**Fig. 4** (a) Macroscopic finding reveals yellow-tan polypoid mass. (b,c) Microscopic findings reveal typical histological appearance of paraganglioma (hematoxylin and eosin stain).

providing the advantage of reducing the risk of a positive surgical margin and ureter or ureteral orifice injury. In addition, using the needle electrode, we first excised circumferentially around the lesion with sufficient margin and resected the drainage vein of the tumor without violating the tumor. Manipulation of the tumor before resection of the drainage vein can induce excessive hormone release, resulting in cardiovascular instability. Therefore, this procedure has the advantage of reducing the risk of the elevation of blood pressure during the operation.

Recently, LECS is reported to be a more feasible and safer minimally invasive treatment for submucosal tumors such as glomus tumors and gastrointestinal stromal tumors; LECS is one of the standard options for the resection of these tumors.<sup>11,12</sup> To our knowledge, this is the first report on the resection of paraganglioma of the bladder by combination of en bloc TUR and laparoscopic partial cystectomy; this combination is minimally invasive and safe. Our novel approach using LECS can be considered as one of the options for the treatment of paraganglioma of the bladder.

## Conflict of interest

The authors declare no conflict of interest.

## Editorial Comment

### Editorial Comment to Combination of en bloc transurethral resection with laparoscopic partial cystectomy for paraganglioma of the bladder

Pheochromocytoma of the urinary bladder is a rare disease. However, urologists occasionally encounter patients with this

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disease, as evidenced by many published case reports. A review of the literature revealed 54% patients with pheochromocytoma had hypertension, 48% had headache, and 47% had hematuria as presenting symptom. Furthermore, micturition attack was reported in 53% of patients.<sup>1</sup> In the absence of clear guidelines, urologists encountering this disease often find it difficult to choose the appropriate treatment option. Transurethral tumor resection, open partial cystectomy, laparoscopic partial cystectomy, and radical cystectomy with