Case Report

A case of robot-assisted adrenalectomy performed for an adrenal tumor (anastomosing hemangioma) exceeding 7 cm

Toshinori Nishikimi, Hideki Mizuno, Ayano Kashima, Hiroko Morikami, Shigeki Ishiguro, Tomoyoshi Ohashi and Hiroshi Yamada

Department of Urology, Japanese Red Cross Aichi Medical Center Nagoya Daini Hospital, Japan

Abbreviations & Acronyms
MRI = magnetic resonance imaging
LA = laparoscopic adrenalectomy
RA = Robot-assisted adrenalectomy

Correspondence: Toshinori Nishikimi M.D., Ph.D., Department of Urology, Japanese Red Cross Aichi Medical Center Nagoya Daini Hospital, 2-9 Myoken-cho, Showa-ku, Nagoya, Aichi 466-8650, Japan. Email: nishiki@nagoya2.jrc.or.jp

How to cite this article: Nishikimi T, Mizuno H, Kashima A *et al*. A case of robot-assisted adrenalectomy performed for an adrenal tumor (anastomosing hemangioma) exceeding 7 cm. *IJU Case Rep*. 2022; 5: 469–473.

Received 29 May 2022; accepted 24 July 2022. Online publication 8 August 2022

Abstract:

Introduction: Anastomosing hemangioma in the adrenal area is extremely rare. We report a large anastomosing hemangioma in the adrenal area that underwent robot-assisted adrenalectomy.

Case presentation: A 49-year-old man with left back pain underwent magnetic resonance imaging (MRI) that revealed a tumor in the left adrenal area; it was diagnosed as nonfunctional endocrinologically. However, the major axis of the tumor increased from 64 to 72 mm during the 4-month period. Robot-assisted left adrenalectomy was performed. Although the large tumor adhered to the surrounding tissues, it was safely resected by the effective use of an extra robotic arm. An anastomosing hemangioma was diagnosed since there were no malignant findings.

Conclusion: Robotic surgical systems may serve as an effective treatment option for large adrenal tumors, and our report is the first robot-assisted adrenalectomy performed on an anastomosing hemangioma.

Key words: adrenal tumor, adrenalectomy, anastomosing hemangioma, robot-assisted surgery, robotic surgical system.

Keynote message

Anastomosing hemangioma in the adrenal area is extremely rare. Robotic surgical system may serve as an effective treatment option for large adrenal tumors; hence, we report the first case of a large anastomosing hemangioma in the adrenal area that underwent robot-assisted adrenalectomy.

Introduction

Anastomosing hemangioma in the adrenal area is extremely rare. We report a large anastomosing hemangioma in the adrenal area that underwent robot-assisted adrenalectomy.

Case

A 49-year-old man with a history of hypertension was admitted to a hospital with a left back pain. When the patient underwent abdominal MRI, a mass was detected in the left adrenal gland; he was then referred to our hospital. Based on the CT (Fig. 1) and MRI findings, no findings were strongly suggestive of malignancy. The hormone levels, in contrast to the tumor diameter, indicated a diagnosis of a nonfunctional tumor. The major axis of the tumor increased from 64 to 72 mm during the 4-month period. Thus, total resection of the tumor, along with the left adrenal gland, was planned. A robot-assisted left adrenalectomy was selected as the surgical procedure. Surgical findings are shown in Figure 2. Histopathological findings were consistent with anastomosing hemangioma (Fig. 3).

Discussion

Robot-assisted adrenalectomy (RA) is performed worldwide. Although laparoscopic adrenalectomy (LA) has been performed as a minimally invasive surgery for adrenal tumors, the effectiveness of RA in comparison with LA remains controversial because of the lack of large-scale randomized prospective studies. In a large-scale meta-analysis of 1,162 patients, a comparison between 747 patients treated with RA and 415 patients treated with LA showed no significant differences in the rates of intraoperative and postoperative complications or mortality; however, the operative time was substantially longer in patients treated with RA.[1] On the effectiveness of RA, Agcaoglu et al.[2] reported that when adrenal tumors exceeding 5 cm were operated, RA was associated with a shorter operative time and fewer cases of conversion to laparotomy than LA. Morelli et al.[3] compared 41 patients treated with RA and 41 patients treated with LA and reported that RA was more effective than LA in patients with tumors measuring ≥6 cm. These reports suggest that RA may be effective for tumors with diameters of 5–6 cm or larger. Piccoli et al.[4] compared 84 patients treated with LA and 76 patients treated with RA and reported that the operative time was longer for RA than for LA when tumors were located on the right side; however, there was no difference in the operative time between RA and LA when tumors were located on the left side. Furthermore, they reported that the incidence of postoperative complications was lower in patients undergoing RA. Favorably, our patient had a left-sided tumor. Despite the large tumor measuring 7.2 cm, the effective use of the extra robotic arm allowed us to complete the procedure smoothly, thereby demonstrating the benefits of RA. The use of an extra robotic arm or sealing device seems effective in RA for adrenal tumors with a large diameter. When the costs of the devices used at our hospital are examined, the cost of RA for tumors of standard size (131,900 yen) is 1.25 times higher than the cost of LA (105,800 yen). When an extra-arm and a sealing device are used in RA for adrenal tumors of large diameters, like the tumor treated in this case, there is a 1.73-fold increase in the cost of RA (183,167 yen). Although RA has advantages, there is also an undeniable problem related to the increased cost of RA due to the use of these devices. Some reports have described the higher cost of RA than that of LA as a problem.[5,6]

Anastomosing hemangioma was first reported by Montgomery and Epstein in six cases in 2009 as a benign tumor with histopathological features similar to those of angiosarcoma.[7] Tao et al.[8] reported that this tumor develops in the kidneys, perinephric fat, liver, ovaries, small intestine, and adrenal gland. Tumors developing in the kidneys and perinephric fat accounted for 61.3% and those developing in the adrenal glands accounted for 3.2%. Most tumors develop in the retroperitoneal region[8] . Perdiki et al.[9] reported that approximately 60% of the patients were asymptomatic. Our

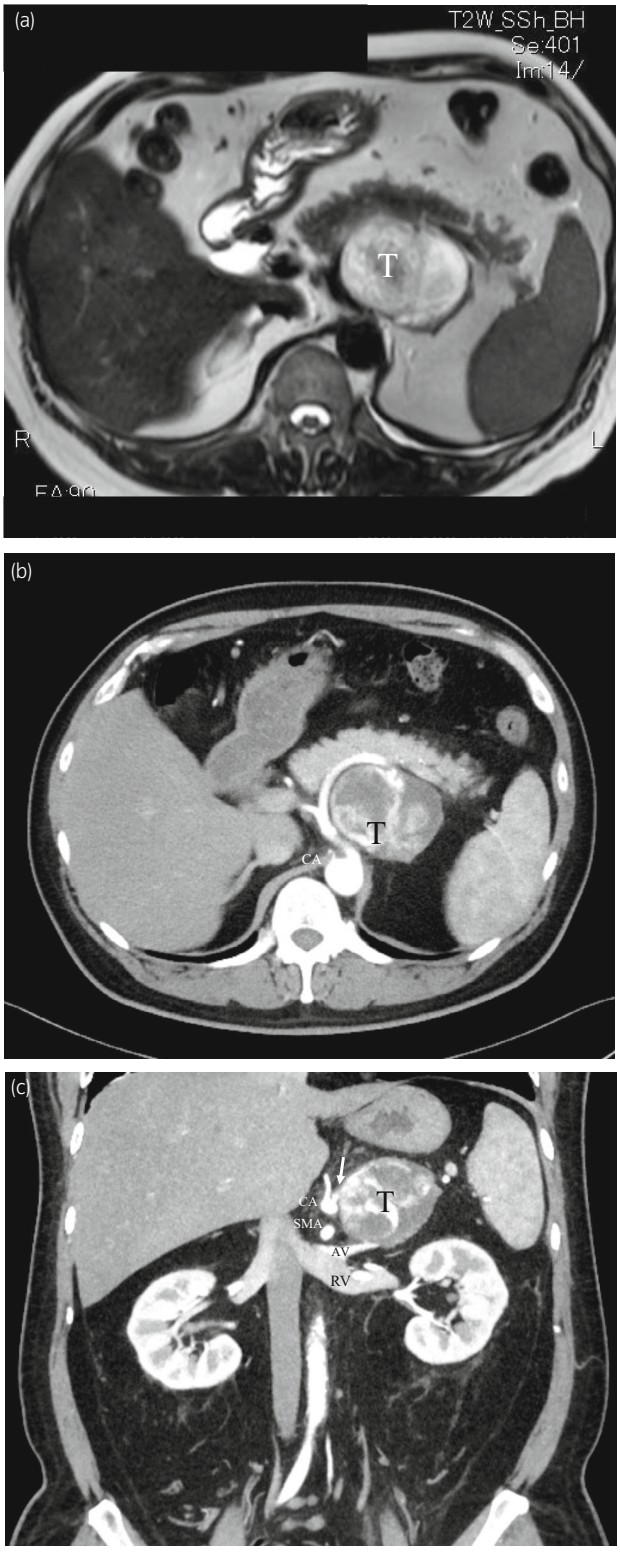

Fig. 1 Imaging findings. (a) Magnetic resonance imaging. A well-demarcated mass measuring 51 × 64 × 56 mm was detected in the left adrenal area. On T2-weighted images, it appeared hyperintense, and the interior signal intensity was heterogeneous. (b, c) Contrast-enhanced abdominal computed tomography. A mass measuring 72 × 64 × 56 mm was detected in the left adrenal gland. It is multinodular and internally heterogeneous. The lesion was enhanced in the early phase of contrast enhancement, and the enhancement was sustained in the late phase. The middle adrenal artery branches from the celiac artery and the adrenal vein drains into the left renal vein. (T, tumor; Av, left adrenal vein; RV, left renal vein; CA, celiac artery; SMA, superior mesenteric artery).

patient presented with left back pain as a chief concern, which might have been caused by compression of the tumor due to its large size. Contrast-enhanced CT shows temporal enhancement starting from the peripheral area to the center of the tumor and MRI depicts this tumor as hyperintense on T2-weighted images.[10] However, diagnosis is difficult based on imaging studies alone.[11] Although anastomosing hemangioma is pathologically similar to angiosarcoma due to a lack of lobular structure, the presence of a pattern of local invasion into blood vessels, hobnail-like endothelial cells, hyaline globules, and dense focal proliferation of blood vessels, it differs from angiosarcoma due to the lack of characteristics indicative of malignancy, such as dyskaryosis, multilayered endothelial cells, and necrotic areas.[10,12]

Our search in PubMed with the keywords, "anastomosing hemangioma" and "adrenal" yielded four case reports.[13–16] In all cases, total resection of the tumor along with the adrenal gland was performed, and the outcome was favorable. No recurrence was reported in any of these cases. Table 1 summarizes the five cases, including ours. Our patient had the largest

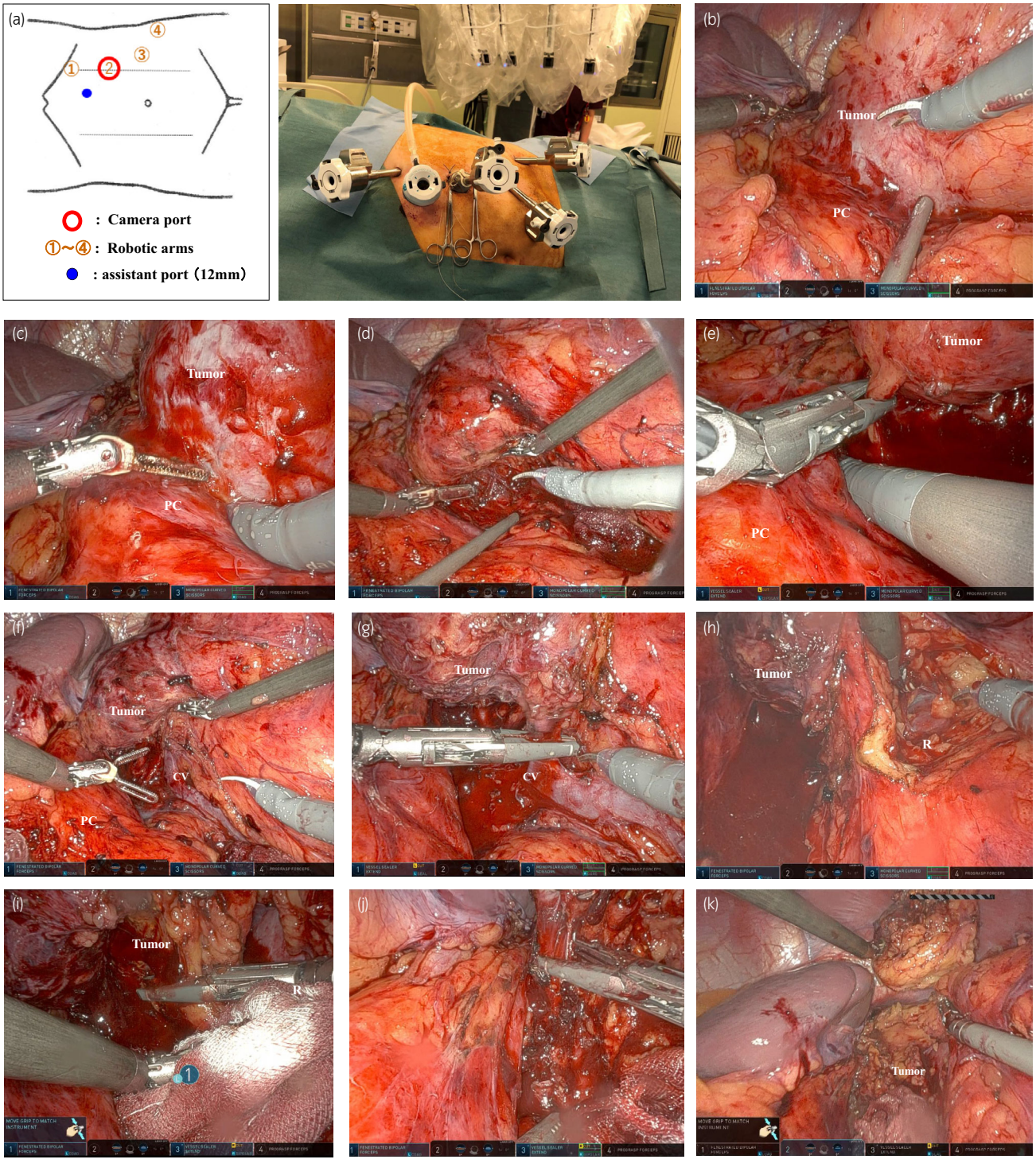

Fig. 2 Surgical findings. (a) Port position. Four robotic arms were docked to the ports. The cranial port for the left robotic arm was placed at the lateral border of the rectus abdominis muscle, closest to the rib. The camera port was placed at the lateral border of the rectus abdominis muscle at a distance of approximately three to four fingers caudally from the port for the left robotic arm. A port for the right robotic arm was placed outside the rectus abdominis muscle, farther from the camera port, also with a distance of approximately three to four fingers. Surgery was initiated using three robotic arms. However, because the tumor was large and exposure of the operative field was difficult, we decided to use an extra robotic arm from a port placed caudally and laterally farther away from the port for the right robotic arm by a distance of approximately three to four fingers. A 12-mm assistant port was placed between the port for the left robotic arm and the camera port. (b) After the lateral borders of the descending colon and spleen were incised to expose the operative field, the tumor was located in the vicinity of the pancreas (PC). (c) Since the tumor adhered to the pancreas, they were carefully dissected. (d) After the tumor was dissected from the pancreas to some extent, we were able to lift the caudal side of the tumor using the extra robotic arm. (e) To further expose the operative field, the medial side of the tumor was dissected. However, because bleeding from around the tumor persisted, in addition to adhesion, a sealing device for robot-assisted surgery (vessel sealer) was used. (f) While the tumor was lifted with an extra robotic arm to expose the operative field, further dissection of the tumor allowed us to identify the central vein (CV) of the adrenal gland. (g) The central vein (CV) was dissected and then resected using a vessel sealer. (h, i) The tumor was dissected from the left kidney. The renal capsule (R) was exposed to avoid incision into the tumor because the poorly demarcated border of the tumor and the hard surrounding fat layer made dissection difficult. The tumor was then dissected at a line that left perinephric fat attached to the adrenal gland (2h). The fat tissue between the dissected left kidney and the tumor were resected with a vessel sealer (i). (j, k) Finally, while the tumor was lifted by grasping the perinephric fat with the extra robotic arm, the medial and cranial sides of the tumor were resected with a vessel sealer (j) to free the tumor (k). After the absence of major bleeding at the resection site was confirmed, the wound was closed to complete the surgery. The operative time was 4 h and 34 min, and the console time was 3 h and 29 min. The blood loss volume was 250 mL. Mobilization and oral nutrition were started the day after surgery, and the patient was discharged from our hospital 4 days after surgery.

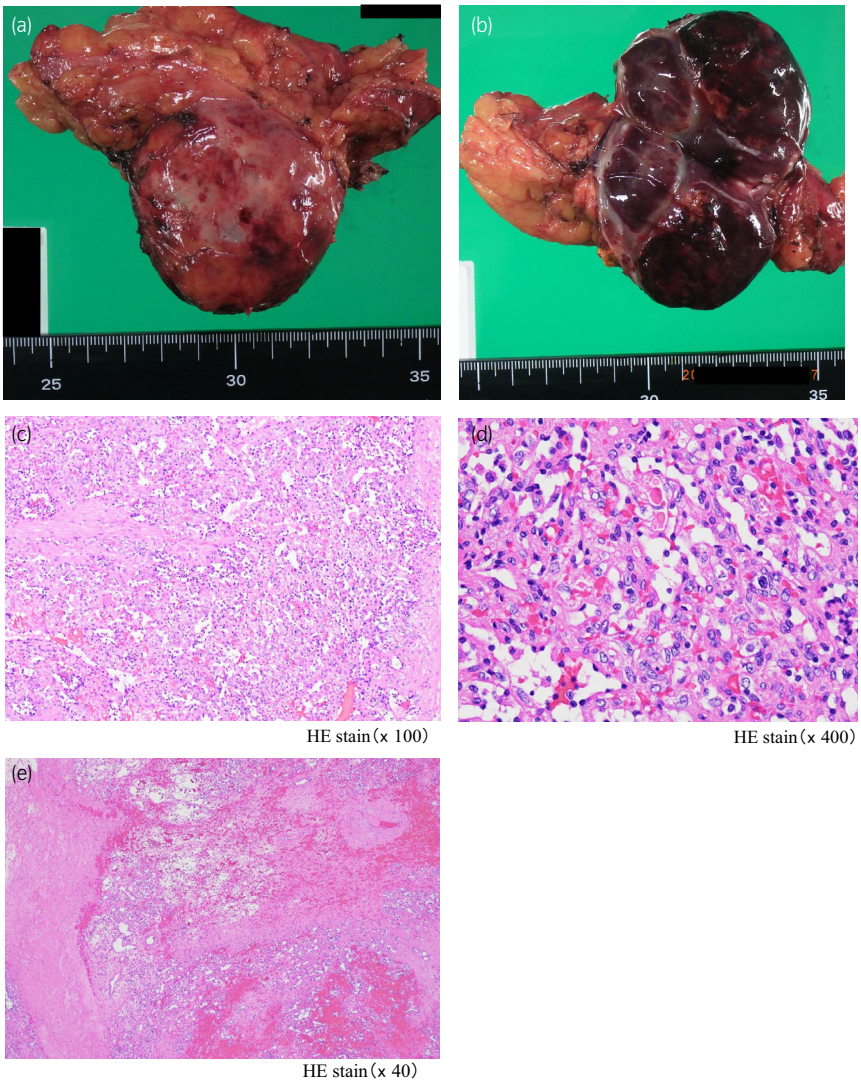

Fig. 3 Histopathological findings. (a, b) Macroscopically, a well-demarcated solid tumor formed in the fat tissue around the left adrenal gland, and the cut surface appeared dark red. The resected tumor weighed 105 g. (c, d) Histologically, small vessels and reticular/anastomotic sinusoids formed a lobular structure and simultaneously proliferated to become compressed. Endothelium-like tumor cells lining the vascular lumens exhibited mild nuclear enlargement and a hobnail-like proliferation pattern (c) and contained scattered hyaline globules (d). An anastomosing hemangioma was diagnosed since there were no malignant findings. (e) Bleeding and edematous changes had occurred in the tumor.

anastomosing hemangioma that developed in the adrenal gland, and our report is the first RA performed on an anastomosing hemangioma. Among endocrinologically inactive adrenal tumors that are accidentally detected, resection is recommended for those measuring ≥4 cm and those suspected to be malignant on imaging.[17] In our case, the tumor exceeded 7 cm on the preoperative images; hence, it was totally resected along with the adrenal gland. Since the intraoperative findings also showed that the tumor firmly adhered to the surrounding tissues as well as to the adrenal gland, we assumed that preservation of the adrenal gland was difficult. In any case, anastomosing hemangioma in the adrenal area is extremely rare, and no consensus has been reached on treatment. Further

Table 1 Case reports of anastomosing hemangioma in adrenal area

	Publication year	Age/Sex	Affected side	Size (in mm)	Symptom	surgical procedure	Another lesion	recurrence
Ross M	2012	49/M	Right	37 × 33	asymptomatic	Laparoscopic radical nephrectomy/ adrenalectomy	None	None
Burton KR	2017	68/M	Bilateral	21: Left (the biggest)	asymptomatic	Laparoscopic left adrenalectomy	Left peri renal fat, right retrocaval, right adrenal,	None
Patel SR	2019	39/M	Right	10	Incidental (acute pericarditis)	nephrectomy/ adrenalectomy	Bilateral kidney	None
Oki Y	2021	83/M	Left	40 × 36	Left back pain	Laparoscopic adrenalectomy	None	None
Our case	2022	69/M	Right	72 × 64 × 56	Left back pain	robot-assisted adrenalectomy	None	None

accumulation of cases will lead to the establishment of diagnostic and therapeutic methods in the future.

Conclusion

We reported a large size of anastomosing hemangioma in the adrenal area that underwent robot-assisted adrenalectomy. Robotic surgical systems may serve as an effective treatment option for large adrenal tumors, and our report is the first robot-assisted adrenalectomy performed on an anastomosing hemangioma.

Acknowledgments

None.

Author contributions

Toshinori Nishikimi: Conceptualization; data curation; formal analysis; investigation; methodology; project administration; resources; software; supervision; validation; visualization; writing – original draft; writing – review and editing. Hideki Mizuno: Methodology; validation. Ayano Kashima: Validation. Hiroko Morikami: Validation. Shigeki Isiguro: Validation. Tomoyoshi Ohashi: Validation. Hiroshi Yamada: Validation.

Conflict of interest

The authors declare no conflicts of interest. In consideration of ethical aspects, the contents of this article comply with the provisions of the ethics committee of the Japanese Red Cross Aichi Medical Center Nagoya Daini Hospital.

Approval of the research protocol by an institutional reviewer board

The protocol for this research project has been approved by a suitably constituted Ethics Committee of the institution. Informed consent was obtained from the subjects.

The registration No.001219.

Registry and the Registration No. of the study/trial

Not applicable.

References

1 Economopoulos KP, Mylonas KS, Stamou AA *et al*. Laparoscopic versus robotic adrenalectomy: A comprehensive meta-analysis. *Int. J. Surg.* 2017; **38**: 95–104.
2 Agcaoglu O, Aliyev S, Karabulut K, Mitchell J, Siperstein A, Berber E. Robotic versus laparoscopic resection of large adrenal tumors. *Ann. Surg. Oncol.* 2012; **19**: 2288–94.
3 Morelli L, Tartaglia D, Bronzoni L *et al*. Robotic assisted versus pure laparoscopic surgery of the adrenal glands: a case-control study comparing surgical techniques. *Langenbecks Arch. Surg.* 2016; **401**: 999–1006.
4 Piccoli M, Pecchini F, Serra F *et al*. Robotic versus laparoscopic adrenalectomy: pluriannual experience in a high-volume center evaluating indications and results. *J. Laparoendsc. Adv. Surg. Tech. A.* 2021; **31**: 375–81.
5 Samreen S, Fluck M, Hunsinger M, Wild J, Shabahang M, Blansfield JA. Laparoscopic versus robotic adrenalectomy: a review of the national inpatient sample. *J. Robot Surg.* 2019; **13**: 69–75.
6 Bodner J, Augustin F, Wykypiel H *et al*. The Da Vinci robotic system for general surgical applications: a critical interim appraisal. *Swiss Med. Wkly.* 2005; **135**: 674–8.
7 Montgomery E, Epstein JI. Anastomosing hemangioma of the genitourinary tract. *Am. J. Surg. Pathol.* 2009; **33**: 1364–9.
8 Tao LL, Dai Y, Yin W, Chen J. A case report of a renal anastomosing hemangioma and a literature review: an unusual variant histologically mimicking angiosarcoma7. *Diagn. Pathol.* 2014; **9**: 159.
9 Perdiki M, Datseri G, Liapis G *et al*. Anastomosing hemangioma: report of two renal case and analysis of the literature. *Diagn. Pathol.* 2017; **12**: 14.
10 Cheon PM, Rebello R, Naqvi A, Popovic S, Bonert M, Kapoor A. Anastomosing hemangioma of the kidney: radiologic and pathologic distinctions of a kidney cancer mimic. *Curr Oncol* 2018; **25**: e220–3.
11 O'Neill AC, Craig JW, Silverman SG *et al*. Anastomosing hemangioma: locations of occurrence, imaging features, and diagnosis with percutaneous biopsy. *Abdom Radiol (NY)* 2016; **41**: 1325–32.
12 Lappa E, Drakos E. Anastomosing hemangioma: short review of a benign mimicker of angiosarcoma. *Arch. Pathol. Lab. Med.* 2020; **144**: 240–4.
13 Patel SR, Abimbola O, Bhamber T, Weida C, Roy O. Incidental finding of bilateral renal and adrenal anastomosing hemangiomas: A rare case report. *Urol Case Rep* 2019; **27**: 100912.
14 Burton KR, Jakate K, Pace KT, Vlachou PA. A case of recurrent, multifocal anastomosing haemangiomas. *BMJ Case Rep.* 2017; **2017**:220076
15 Ross M, Polcari A, Picken M, Sankary H, Milner J. Anastomosing hemangioma arising from the adrenal gland. *Urology* 2012; **80**: e27–8.
16 Yuta Oki Y, Sato M, Horitani H *et al*. A case of adrenal anastomosing hemangioma. *Hinyoukika Kiyo* 2021; **67**; 7–10
17 Berland LL, Silverman SG, Gore RM *et al*. Managing incidental findings on abdominal CT: white paper of the ACR incidental findings committee. *J. Am. Coll. Radiol.* 2010; **7**: 754–73.

