

Robotic treatment of giant adrenal myelolipoma: A case report and review of the literature

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Abstract. Adrenal myelolipoma is a rare mesenchymal tumour with benign biological behaviour that is mainly composed of mature adipose and myeloid tissue. Both sexes are equally affected, most commonly between the fifth and seventh decades of life. The diagnosis of adrenal myelolipoma is mostly incidental. Although it may occasionally be associated with necrosis, rupture and haemorrhage, causing abdominal pain, this tumour is usually asymptomatic. Consequently, management is conservative, while surgical treatment is reserved for symptomatic cases, or for masses growing quickly or to a size >6 cm. Giant myelolipomas (sized >10 cm) are rare. Open radical adrenalectomy is the standard treatment for giant myelolipomas, while the minimally invasive approach has been used in only few cases. We herein report the case of a patient with a giant adrenal myelolipoma who underwent robotic partial adrenalectomy. To the best of our knowledge, this is the largest giant adrenal myelolipoma treated with robotic surgery reported in the literature to date. A 55-year-old male patient underwent an abdominal computed tomography scan during follow-up after radical prostatectomy for prostate cancer Gleason Score 6 (ISUP 1) due to biochemical recurrence. The examination revealed a right hypodense adrenal mass, sized 16x13 cm. Abdominal magnetic resonance imaging confirmed the presence of characteristics suggestive of a myelolipoma. The patient did not report any symptoms. Due to the benign characteristics of the mass, robotic partial

adrenalectomy and enucleation of the mass were performed. The operative time and estimated blood loss were 205 min and 100 ml, respectively. No intra- or postoperative complications occurred. The patient was mobilized on the first postoperative day and the time to flatus was 36 h; the length of hospitalization was 4 days. Histological examination confirmed the diagnosis of adrenal myelolipoma, sized 18x11.5x6 cm. No tumour recurrence occurred over a follow-up period of 12 months. In conclusion, robotic surgery allows performing partial adrenalectomy with a lower risk of bleeding and with preservation of healthy adrenal tissue, which is of paramount importance for the patient as it reduces recovery time and the need for medical substitution therapy.

Introduction

Adrenal myelolipoma is a rare mesenchymal tumour with benign biological behaviour that is mainly composed of mature adipose and myeloid tissue (1). Its pathogenesis is largely unknown, but infection, inflammation, necrosis, stressful lifestyle and an unbalanced diet are known risk factors; adrenal myelolipoma is also often associated with obesity, high levels of blood lipids, hypertension, diabetes and Cushing's disease (2). The two sexes are equally affected, mainly between the fifth and seventh decades of life (3). The most common site is the right adrenal gland (4), but other sites, such as the presacral area, spleen, stomach, lung, liver, retroperitoneum and testis, are also reported (5).

Adrenal myelolipoma is generally non-secretory and its diagnosis is most commonly incidental: It currently represents 10-15% of incidental adrenal masses discovered on imaging investigations, such as ultrasonography, computed tomography (CT) and magnetic resonance imaging (MRI). Myelolipoma is usually asymptomatic, but it may occasionally be associated with necrosis, rupture and haemorrhage, causing abdominal pain (6). Due to the absence of symptoms and its small size (usually <4 cm), the management of myelolipoma is usually conservative. Surgical treatment is recommended when the mass is symptomatic, or grows quickly or to a size of >6 cm (6). Adrenalectomy must be performed in case of malignant or potentially malignant tumors (1). Myelolipoma is defined as 'giant' when its greatest diameter is >10 cm (2), which is a rare clinical occurrence. Open radical surgery is

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Abbreviations: CT, computed tomography; MRI, magnetic resonance imaging; OT, operative time; EBL, estimated blood loss

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the standard treatment of choice for giant myelolipomas, while the minimally invasive approach has been used in only a few cases (7). We herein present the case of a patient with an 18-cm adrenal myelolipoma who was treated by robotic partial adrenalectomy. To the best of our knowledge, this is the largest adrenal myelolipoma treated with robotic surgery reported in the literature to date.

Case report

A 55-year-old male patient underwent an abdominal CT scan during follow-up after radical prostatectomy. The examination revealed a right adrenal mass, sized 16x13 cm, homogeneous, hypodense (<30 HU), imprinting on the right liver lobe and dislocating the right kidney downwards, the vena cava, duodenum and pancreas medially, and the ascending colon and gallbladder anteriorly. Following administration of contrast medium, the adrenal lesion exhibited poor enhancement with a rapid washout. The abdominal MRI confirmed the presence of a right adrenal lesion, sized 16x13 cm. Hyperintense on T1-weighted, isointense on T2-weighted and hypointense on fat-suppression images, without significant contrast enhancement, characteristics suggestive of a myelolipoma (Fig. 1). The patient did not report any symptoms, but exhibited a deformation of the right abdominal contour. Staging workup excluded other adrenal diseases.

The patient did not have other major comorbidities, and had a Charlson Comorbidity index score of 3 and an Eastern Cooperative Oncology Group performance status score of 0.

Due to the benign characteristics of the mass, it was decided to perform transperitoneal robotic partial adrenalectomy in order to preserve adrenal healthy tissue and maintain functional adrenal integrity in a young patient undergoing surgery for a benign lesion.

Written informed consent was obtained from the patient prior to surgery.

The operation was performed by a robotic-skilled urologist, using the DaVinci® Model Xi Surgical System (Intuitive Surgical, Inc., Sunnyvale, CA, USA) in the Tertiary Care Hospital Santa Maria di Terni (Terni, Italy). The patient was placed in a left flank position with a 45° side tilt. The optical trocar was inserted on the intersection between the transumbilical and periumbilical lines. Two more robotic trocars and two trocars for the assistant were inserted (Fig. 2). The triangular hepatic ligament was dissected to mobilize the liver, followed by enucleation of the mass (Fig. 3). The operative time (OT) and estimated blood loss (EBL) were 205 min and 100 ml, respectively. No intra- or postoperative complications occurred. On the first postoperative day, the nasogastric tube was removed and the patient was mobilized; the time to flatus was 36 h, oral feeding was initiated 3 days after surgery and the length of hospitalization was 4 days. The histological examination confirmed the diagnosis of adrenal myelolipoma, sized 18x11.5x6 cm. Follow-up examinations included an abdominal ultrasound and CT scan at 6 and 12 months after surgery, respectively. At 12 months of follow-up no recurrence had occurred.

A systematic bibliography search up to January 2018 was conducted through PubMed, Web of Science and Scopus. One of the authors (AP) independently performed online

bibliographic research in order to identify titles and abstracts of interest. All titles and abstracts were assessed to select those focusing on minimally invasive adrenalectomy for sizeable masses. Subsequently, the full text of the selected articles was independently screened by two authors (ES and JARV) for eligibility (Table I).

Discussion

Adrenal myelolipoma is a rare benign tumour with a prevalence of 0.08-0.2% (3) that consists of mature adipose and hematopoietic tissue (8). Myelolipoma was first described by Gierke in 1905, and was named 'formations myelolipomatoses' by Oberling in 1929 (9). In the majority of the cases, it is incidentally diagnosed during clinical workup for other reasons. Myelolipoma mainly presents with abdominal discomfort and pain due to necrosis, rupture, haemorrhage, or even haemorrhagic shock (6). The mean size at the time of diagnosis is ~4 cm, while myelolipomas >10 cm in greatest diameter are defined as 'giant' (2). The management of smaller, asymptomatic, slow-growing lesions is conservative, and patients are monitored by abdominal CT or MRI annually or biannually (2). Surgical treatment is recommended for asymptomatic tumours >6 cm in diameter, for symptomatic lesions and for those exhibiting fast growth (6). In the AACE/AAES Adrenal Incidentaloma Guidelines, resection of myelolipoma may be considered under certain conditions, such as the presence of clinical symptoms or a diameter of >4 cm (10). Based on the preoperative imaging examinations, partial adrenalectomy was considered to be the best choice in the present case.

An open or a minimally invasive approach is selected according to the size of the lesion and the skills of the surgeon. The open approach includes lumbar, subcostal, posterior or transabdominal laparotomy, and is usually reserved for giant masses or as an emergency intervention in cases with rupture and haemorrhage.

The advantages of the laparoscopic compared with the traditional approach are less pain, faster recovery and shorter hospitalization, and it is usually reserved for smaller myelolipomas (11,12). Robotic surgery is being increasingly used in different common surgical procedures, although its applicability in adrenal surgery remains debated upon (13,14). A magnified 3D view of the operative field, the absence of tremor, wristed instruments and improved ergonomics are considered invaluable advantages over the laparoscopic approach.

Giant myelolipomas are associated with more intraoperative complications, such as bleeding, capsular breach and a higher risk of local recurrence (9,15). Morelli *et al* reported better results with the robotic vs. the laparoscopic approach in patients with large adrenal tumours (>6 cm in diameter). The OT for lesions sized >6 cm was shorter in the robotic group (171.3 vs. 260.0 min, P=0.002). There were no significant differences between the two groups regarding the rate of conversion, intra- and postoperative complications (P=0.49, 0.95 and 0.96, respectively) (16).

However, only few cases of minimally invasive adrenalectomy for giant adrenal myelolipomas have been reported to date (Table I). Campos Arbulú *et al* described a laparoscopic adrenalectomy for a 14-cm myelolipoma; no complications occurred and the length of hospitalization was 48 h (17).

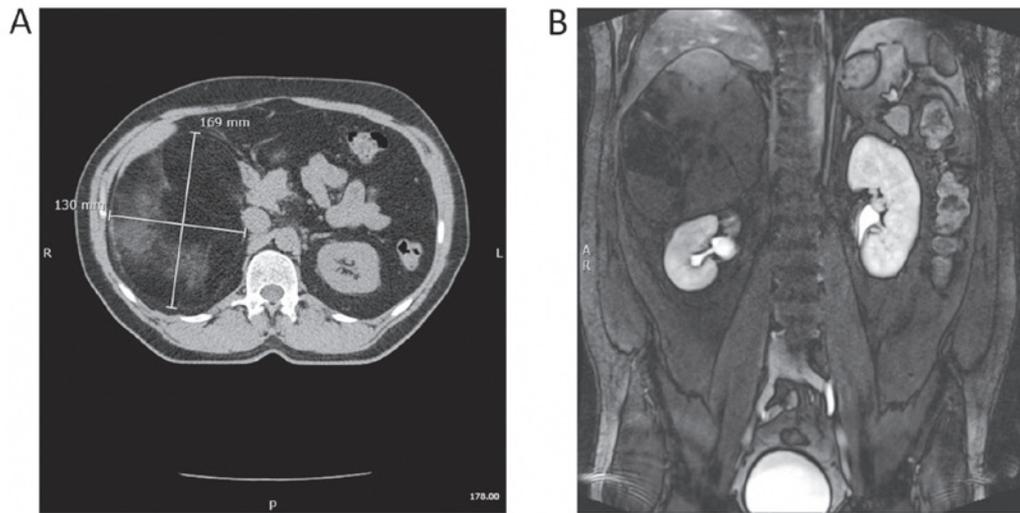


Figure 1. (A) Computed tomography and (B) magnetic resonance imaging scan showing the giant right adrenal myelolipoma.

- Optical trocar 8 mm (O)
- Robotic trocar 8 mm (R)
- AirSeal® 10-12 mm (AS)
- Liver retractor 5 mm (L)

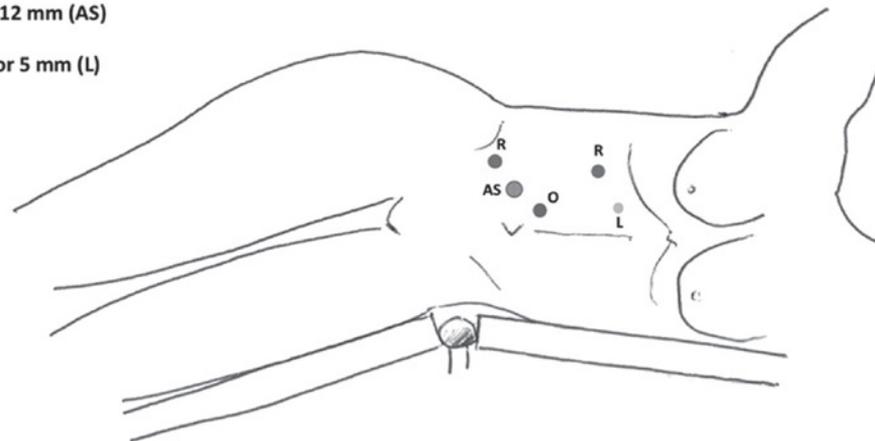


Figure 2. Trocar placement. The optical trocar (O) was positioned between the transumbilical and periumbilical lines. Two robotic trocars (R) were placed on the pararectal line at 8 cm from O. The AirSeal® trocar (AS) was placed between O and the more caudal R. The liver retractor trocar (L) was placed subxifoidally on the paraumbilical line.

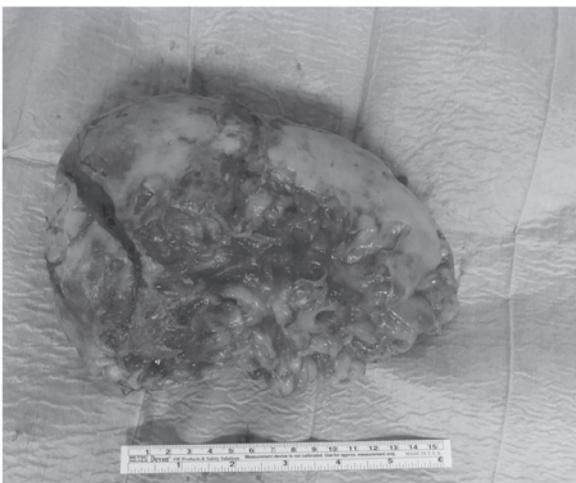


Figure 3. Macroscopic view of the resected giant myelolipoma.

Undre *et al* performed robotic adrenalectomy for a 13.9-cm adrenal myelolipoma. The OT was 155 min and the EBL was 300 ml. No complications occurred and the length of hospitalization was 4 days (18). Brunaud *et al* performed 100 robotic adrenalectomies; tumour capsule rupture occurred in only 1 patient (1%) with a 7.5-cm lesion (19). There was 1 case of conversion to laparoscopy due to camera malfunction, in 4 cases open conversion was necessary due to bleeding and difficulty to identify the adrenal vein in 1 patient with a 7-cm mass. Of note, all open conversions occurred in patients with high BMI (≥ 29 kg/m², 75%) and large tumours (≥ 6 cm, 50%) (15). Agcaoglu *et al* performed 24 robotic and 38 laparoscopic adrenalectomies. Tumour size was similar in both groups (6.5 ± 0.4 , range 5-10.2 cm for the robotic group; 6.2 ± 0.3 , range 5-15 cm for the laparoscopic group) as were age and sex. The robotic group had lower BMI (27.1 ± 0.8 vs. 30.2 ± 0.9 kg/m²). The robotic approach was associated with a shorter OT (159 vs. 187 min;

Table I. Cases of mini-invasive surgery for adrenal masses.

Study ID	Year	Technique	No. of patients	Tumour size (cm)	Refs.
Molnar, <i>et al</i>	2017	R	1	4.6x4.1	(5)
Economopoulos, <i>et al</i>	2017	L/R	415/353	2.8-6.2/2.57-6.5 (mean)	(21)
Campos Arbulù, <i>et al</i>	2017	L	1	14	(17)
Burtet, <i>et al</i>	2017	R	1	8.3	(22)
Morelli, <i>et al</i>	2016	L/R	41/41	4.7/4.9 (mean)	(16)
Teo and Lim	2016	L/R	263/569	1-15/1-13.9	(7)
Deniwar, <i>et al</i>	2015	R	1	3.9	(23)
Akarsu, <i>et al</i>	2014	R	8	2-9	(24)
Brandao, <i>et al</i>	2013	L/R	323/277	3.78±1.06-3.86±1.32	(25)
Yates, <i>et al</i>	2010	R	1	1.5	(26)

R, robotic; L, laparoscopic.

P=0.043) and lower EBL (87 vs. 167 ml; P=0.147). The conversion rate was markedly higher in the laparoscopic group (11 vs. 4%) and the causes of conversion in the laparoscopic group included bleeding from the renal vein or the adrenal gland, adhesion to the vena cava and difficulty with the dissection plane; in the robotic group, the main cause of conversion was the adherence of the tumour to the renal hilum (20). To the best of our knowledge, the case presented herein is the largest giant myelolipoma treated by robotic approach that has been reported in the literature to date.

A robotic partial adrenalectomy was performed, with enucleation of the mass. No intra- or postoperative complications occurred. The EBL was 100 ml, which was comparable with previous reported cases (7,21,25). The choice of partial adrenalectomy for a benign tumour was based on finding an avascular plane between the tumour capsule and the healthy surrounding adrenal tissue, in order to significantly reduce intraoperative bleeding and preserve healthy adrenal tissue. The robotic approach allowed a safer preservation of the adrenal vessels due to the magnified 3D vision and the wristed instruments that permitted more accurate dissection. Furthermore, no capsular effraction was observed. Several studies demonstrated the feasibility and safety of robotic partial adrenalectomy, with satisfactory oncological as well as functional results (9). In the present case, the functional outcome was optimal, as the patient did not require steroid supplementation.

In conclusion, the standard treatment for giant myelolipoma is laparotomic adrenalectomy. However, while the minimally invasive techniques were initially used for smaller lesions, the improved laparoscopic and robotic instrumentations and the increasing experience of the surgeons have led to the wider application of the minimally invasive approach in more complex cases.

We consider the transperitoneal robotic approach to be a safe strategy for giant adrenal masses due to the better visualization of the operative field and more accurate dissection, decreasing the risk of capsular effraction. Moreover, the enucleation of the mass decreases the risk of bleeding, which is the most common cause of conversion, and enables preservation of adrenal functional healthy tissue.

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Availability of data and materials

All the necessary information on the present case report is provided in this manuscript.

Authors' contributions

AP independently performed online bibliographic research in order to identify titles and abstracts of interest. GC and EM were responsible for the conception and design. AB, GC, ES, AT and JDV participated in either drafting or critically revising the article for important intellectual content. All authors have approved the final version of the manuscript to be published and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Ethics approval and consent to participate

The study has been approved by an Internal Board Committee and written informed consent was obtained from the patient prior to surgery.

Patient consent for publication

The patient provided written consent regarding the publication of the case details and associated images.

Competing interests

The authors declare that they have no competing interests to disclose.

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