Laparoscopic Adrenalectomy in Large Localized Adrenocortical Carcinoma: Case Report and Review of Literature

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ABSTRACT

Introduction and Objectives. Adrenocortical carcinoma is a rare, but aggressive tumour, with heterogeneous biological behavior. Complete surgical resection remains the cornerstone of the treatment for localized disease. The role of laparoscopic approach, the gold standard for benign adrenal diseases, is still controversial for malignant adrenal tumors. The objective of this study was to present a case report of laparoscopic adrenalectomy for a large localized adrenocortical carcinoma.

Materials and methods. We report the case of a 39-year-old female diagnosed with a large adrenal tumour secreting cortisol and androgens that was referred in our service for surgical treatment. The Computed Tomography revealed a large, solid, heterogenous adrenal mass of 11 / 8.5 cm without any imaging suspicion of local invasion of the adjacent structures. After obtaining informed consent, a 3D transperitoneal laparoscopic right adrenalectomy was performed.

Results. No surgical or anesthesiologic complications were encountered during the surgery which was accomplished laparoscopically. The tumour was completely excised, without accidental capsule incision or rupture. Total operative time was 120 minutes. Total blood loss was 200 mL. The patient was discharged in the postoperative day 5. Pathological result revealed high grade pT2NxMxL1V1 R0 adrenocortical carcinoma.
Conclusions. Laparoscopic adrenalectomy for large localized adrenocortical carcinoma remains a challenging, but feasible surgical option in carefully selected patients.

Keywords: adrenal tumour, adrenocortical carcinoma, laparoscopic adrenalectomy, minimally invasive surgery

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INTRODUCTION AND OBJECTIVES

Adrenocortical carcinoma (ACC) is a rare but aggressive malignancy arising from the adrenal cortex, with an annual incidence of 0.5–2 cases per million population [1]. It often presents diagnostic and therapeutic challenges due to heterogeneous clinical manifestations, late presentation, and variable biological behavior [2].

Most of ACCs are sporadic and incidentally diagnosed. They accounted for 1.4% of a large number of incidentalomas in a large Japanese study [3]. The risk of malignancy increases with the tumour size (<4 cm vs > 4 cm). Most of them (60%) are functioning tumours and may secrete any combination of steroid hormones [4].

For adequate imaging characterization, contrast-enhanced CT or MRI is usually performed. ACCs are usually large (>6 cm), heterogeneous, irregular tumours that may contain hemorrhage, necrosis, calcifications [2].

Surgical resection remains the cornerstone of treatment for localized ACC, as complete tumor removal is the most important prognostic factor for long-term survival. However, the optimal surgical approach continues to be debated, especially for large tumors [5].

Laparoscopic adrenalectomy (LA) has become the preferred technique for benign and small adrenal tumors due to its minimal invasiveness, reduced perioperative morbidity, and faster recovery. However, its role in the cases of large adrenal tumours or suspected malignancies, such as ACC, remains controversial. The main concerns regarding incomplete resection and tumor spillage often lead to the selection of open adrenalectomy (OA) for most of such cases. Recent advancements in laparoscopic techniques and equipment, as well as the accumulation of surgical expertise, have expanded the indications for LA, even in challenging scenarios such as large localized ACC [2,4,5,6].

The objective of this study was to present a case report of laparoscopic transperitoneal adrenalectomy for a large localized adrenocortical carcinoma.

MATERIALS AND METHODS

We report the case of a 39-year-old female diagnosed with a large adrenal tumour secreting cortisol and androgens that was referred in our service for surgical treatment.

The patient complains of progressive dyspnea on exertion (then dyspnea at rest), cough with pink-colored expectoration and arterial hypertension. Clinical examination revealed grade I obesity, red "moon face" with hirsutism, generalized peripheral edema and severe arterial hypertension. The computed tomography (CT) of thorax performed for pulmonary symptoms revealed multiple bilateral small pulmonary nodules of variable dimensions, some of them with tendency to consolidation, bilateral hilar and mediastinal lymph nodes enlargement. Along these, a large adrenal mass of 11 / 8.5 cm was described as a solid, heterogenous tumour with regular borders, progressive contrast enhancement, and several areas of necrosis. There was no any suspicion of imaging sign of local invasion of the adjacent organs. The pulmonary lesions were in regression at cross-sectional imaging performed several weeks after finishing the antibiotics prescribed by pneumologist, suggesting an infectious process.

The specific hormonal evaluation performed in the endocrinology department revealed high level of cortisol and androgens hormones (low ACTH < 1.5 pg/mL, late night serum cortisol – 12 ug/dL, late night salivary cortisol – 0.4 ug/dL, free urine cortisol – 126 ug/24h, 1 mg dexamethasone suppression test – positive, serum DHEA-S – 5.35 ug/mL, serum testosterone – 0.65 ng/mL).

All surgical options were discussed with the patient. After obtaining informed consent, a transperitoneal laparoscopic approach was performed.

Surgical technique

After orotracheal intubation, we placed the patient in left lateral decubitus (about 60° tilt) with the table slightly flexed at the level of the umbilicus. We placed the first (optical) trocar of 10 mm using

Hasson technique superior and lateral to the umbilicus, along the lateral border of the rectus abdominis muscle, approximately 5 cm caudal to the costal margin. The second and third trocar were placed under visual control, one (10 mm) at the lateral border of the rectus abdominis muscle, about two fingerbreadths caudal to the costal margin and another (10 mm) in anterior axillary line, between anterosuperior iliac spine and camera port, respectively. In this case, we also put a 5-mm port below xiphoid process for liver retraction, and a 10-mm port just lateral to the anterior axillary line, two fingerbreadths below the costal margin. The two most lateral trocars were alternated between left hand of the surgeon and the assistant.

Any adhesions between the liver and peritoneum, the hepatocolic ligament and the triangular ligament were released in order to lift the liver as cranially as possible using the subxiphoid 5-mm port. The colon and duodenum were medially dissected as much as needed. Open the peritoneum at along the liver, caudally along the vena cava up to the renal vein, and laterally along the diaphragm to mobilize medially the right hepatic lobe (Figure 1).

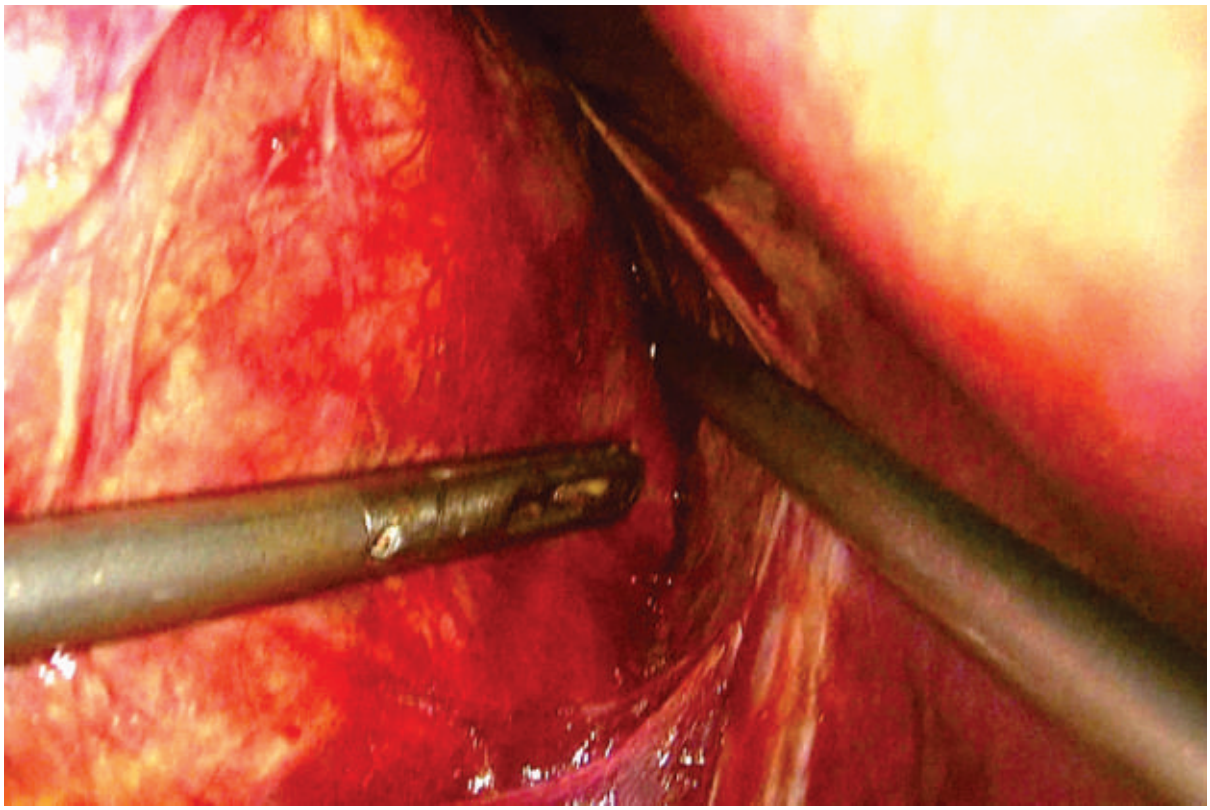


Figure 1. Dissection in the paucivascular plane between adrenal tumour and liver

Then, we proceeded with the dissection of the upper pole of the kidney. We pushed down the kidney, and we dissected dorsal part of the tumour along the posterior parietal muscles, as much as it was possible (Figure 2).

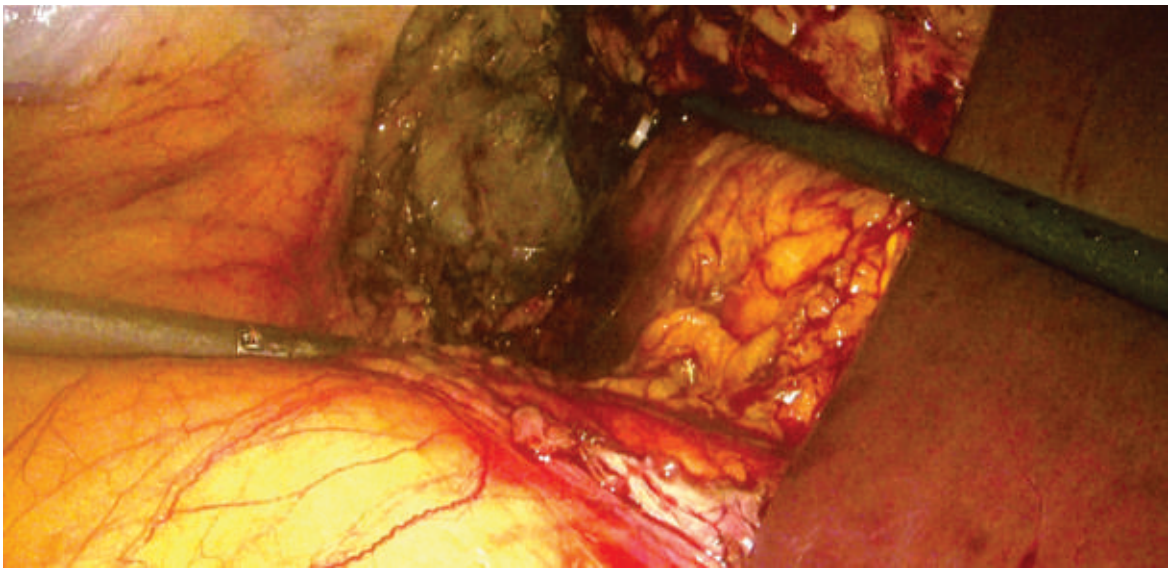


Figure 2. Dissection of the adrenal tumour from the upper pole of the right kidney and parietal muscles

The tumour was lifted and the medial and inferior arterial supply and the adrenal vein were secured using ultrasonic energy and Hem-o-lok clips (Figure 3 and Figure 4).

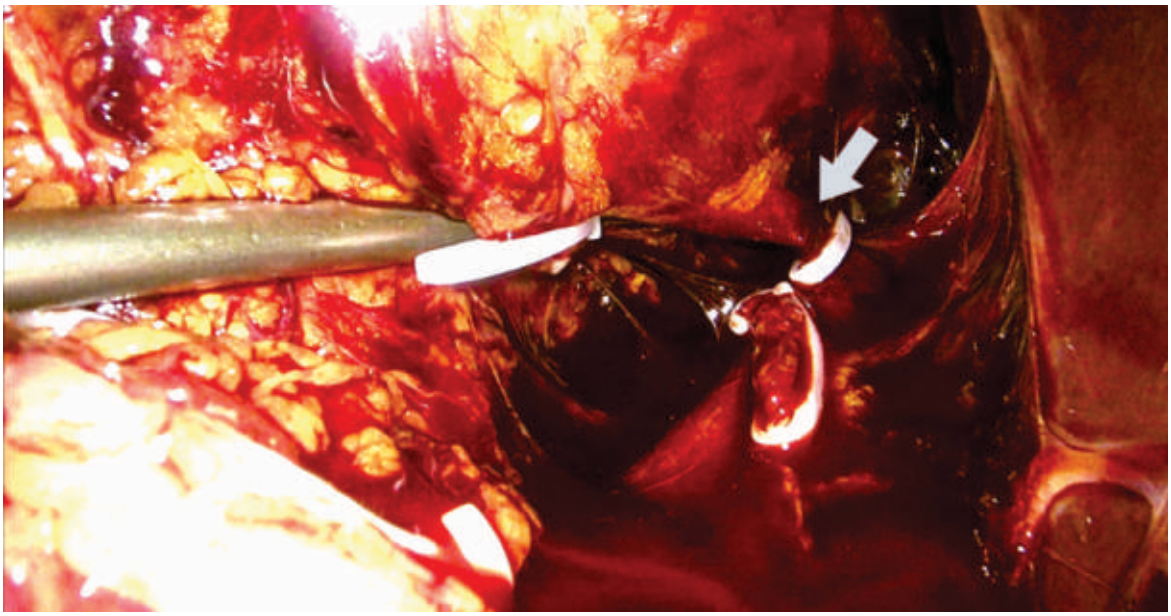


Figure 3. The right adrenal vein clipped (white arrow)

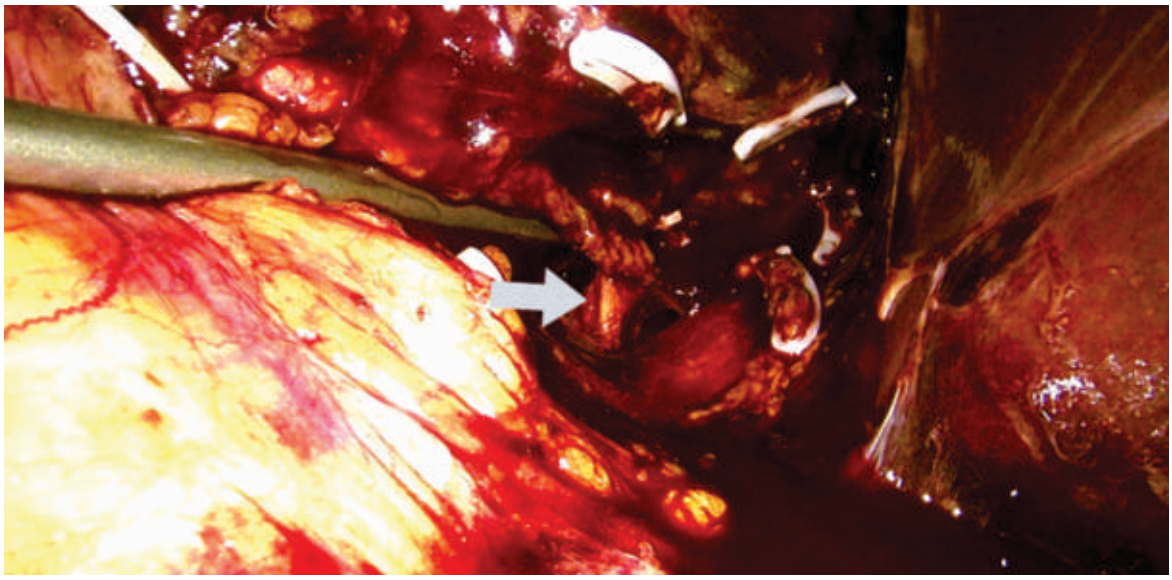


Figure 4. Arterial branch (white arrow) arising from the right renal artery

The circumferential dissection of the tumour was performed, trying to minimize the manipulation of the tumour and to stay away from its capsule. A drainage tube was placed in the adrenal bed. The specimen was placed in an organ bag and removed via extension of the most inferior 10 mm port site. The patient provided her informed consent for participation in our study.

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RESULTS

No surgical or anesthesiologic complications were encountered during the surgery which was accomplished laparoscopically. The tumour was completely excised, without accidental capsule incision or rupture. Total operative time was 120 minutes. Total blood loss was 200 mL. The drain tube was removed in postoperative day 2. The glucocorticoid substitution begun in the afternoon of the surgery, according to the endocrinologist indications. The arterial hypertension begun to be better controlled after surgery. The patient was discharged in the postoperative day 5. The patient was followed regularly by endocrinologist after discharge.

Pathological result revealed high grade pT2NxMxL1V1 R0 adrenocortical carcinoma, with severe cellular and nuclear pleomorphism, with intense mitotic activity, features of necrosis and hemorrhage, isolated sarcomatoid differentiation, and lymphovascular invasion. The patient was referred in oncology department to establish the opportunity of adjuvant chemotherapy and follow up.

DISCUSSION

This case report aimed to contribute to the growing body of evidence on the feasibility, safety, and outcomes of LA in large localized adrenal carcinomas. By presenting the detailed procedure of laparoscopic resection of a large ACC, we highlighted the technical considerations, potential advantages, and limitations of this approach. Additionally, a narrative review of the literature is provided to contextualize our findings and assess the evolving role of minimally invasive surgery in the management of adrenal malignancies.

Understanding the indications and outcomes of LA in such complex cases is critical for optimizing patient care and expanding the surgical options available for managing ACC while maintaining oncological safety [2,5].

Since 1992 when the first laparoscopic adrenalectomy has been performed, this approach experienced an increasing popularity among urologists [4,7]. Laparoscopic approach provides a better visualization and understanding of the anatomy, and is associated with lower postoperative complication rates, including less blood loss and need for transfusion, less postoperative pain and ileus, earlier return to daily activity, better cosmesis, shorter hospital stays, and lower overall costs, when compared with open adrenalectomy. Nowadays, LA is the standard of care for benign, relatively small adrenal tumours [5,7].

Along with technical advancements and increasing expertise of the surgeons in laparoscopy, its indications in adrenal gland surgery were extended, even for ACCs in selected cases. The role of minimally-invasive surgery in the treatment of this malignancy is still under debate. The main concerns are related to the apparently increased risk of capsule rupture, tumour spread in the intraperitoneal space, port site and distant metastasis [2,5,7]. In our case, there were several preoperative imaging and biochemical elements predicting the malignant nature of the tumour: the large diameter (11 cm), presence of necrosis, with heterogeneity, and co-secreting of cortisol and sex hormones.

The size of the tumour is one of the most predictive variables for malignancy. The risk is about 2% for lesions less than 4 cm, whereas for those of 4–6 cm is 6%, and for lesions of 6 cm or higher is 25% (10%–53%), respectively. According to the international guidelines, laparoscopic approach should be considered for small ACCs without signs of loco-regional invasiveness. Although there are not many reports of patients with large malignant tumour, existing data suggest that such patients may undergo LA in the absence of the loco-regional invasiveness. The overall morbidity and mortality are independent of the size of the tumor, and the mean operative time is not significantly different [8,9]. Center volume and the experience of the surgeon seems to be at least as important as the tumour size. The last guidelines recommend that adrenal cancer surgery should be addressed only in centers performing at least six adrenalectomies per year (regardless of the type of approach) and where surgeons are adequately trained in both open and laparoscopic surgery [2,9]. In our patient, the surgeon had adequate expertise in the field of both open and laparoscopic urological surgery, with at least eight adrenalectomies per year.

Although there are comparative studies demonstrating that LA can achieve oncological outcomes comparable to OA, when performed by experienced surgeons, the selection of surgical approach should be individualized, considering factors such as tumor size, location, and the degree of suspicion for malignancy. When there is evidence of local invasion, an open approach should be considered to ensure complete tumor removal and optimal oncological outcomes [2,5]. A retrospective comparative studies involving patients with adrenal masses larger than 6 cm reported that LA resulted in more favorable perioperative results, including shorter operative times, less intraoperative blood loss, and reduced hospital stays compared to OA [5].

Most of the published papers comparing the laparoscopic and open adrenalectomy in ACC included few patients with limited follow-up and methodological variations, thus difficult to extract strong evidence [6]. The results published by two European referral centers on this topic are promising [10,11].

In the study published by Brix et al., 152 patients with localized ACC were included (35 in LA group and 117 in OA group), after a median follow-up of 39.3 months, using both matched pairs approach and multivariate analysis for comparison. The two groups did not differ with regard to the disease-specific survival using either comparison method. The recurrence rate was 77% in the LA group and 69% in the OA group (p=0.36), with a death rate of 37% and 41%, respectively (p=0.68). The adjusted recurrence-free survival was not different between groups (HR: 0.91; 95% CI, 0.56-1.47; p=0.69) [10].
In other study, Porpiglia et al. retrospectively analyzed 43 patients with stages 1 and 2 ACC undergoing radical surgery (25 patients in "open group" and 18 patients in "lap group"), with a

median follow-up of 35 months. They reported a recurrence rate of 64% in the "open group" and 50% in the "lap group". After 3-year follow-up, 84% of the patients for the first group and 100% in the other group were alive (p=0.3). In the multivariate analysis, surgical approach was not an independent prognostic factor of recurrence-free survival [11].

Therefore, more important than concentrating on the choice between laparoscopic and open surgery is the attention on careful manipulation (ACCs are soft and friable tumours), including removal of the tumor mass en bloc with the surrounding fat tissue because of the risk of capsule disruption and tumor cell dissemination. Although the actual coagulation surgical instruments, such as ultrasonic devices or bipolar forceps, demonstrated reducing blood loss, they should be judiciously used. The R0 resection status is the most powerful predictor to achieving long-term survival in patients with ACC. Conversion to open approach should be immediately performed if there are concerns regarding oncological safety. It seems that conversion is not based on tumor size but rather on the difficulty of separating the tumor from the surrounding tissues. Thus, what determines the success of laparoscopic adrenalectomy is not tumor size, but the condition of the tumor, including adhesions and invasion of the adjacent structures. In our patient, the tumour had a well-defined capsule, without imaging and intraoperative suspicion of local invasion [2,6,12].

Performing LA on large adrenal tumors often requires knowledge of several technical tricks changing the sequence of standard surgical steps, as it was shown in our case. The primary difficulty is the limited working space within the abdominal cavity when dealing with large masses. This constraint can hinder instrument maneuverability and visualization. To address this issue, establishing a sufficient pneumoperitoneum and utilizing appropriate patient positioning can enhance exposure and facilitate surgical manipulation [12,13].

Another challenge is the difficult control of the adrenal vein and the arterial branches, which increase the risk of intraoperative bleeding. Early identification and control of the adrenal vein is not always possible as in adrenalectomies for small tumours. As shown in our case, after mobilizing the liver exposure of the inferior vena cava, we proceeded with dissection in the plane between tumour and the upper pole of the kidney, then releasing the dorsal part of the tumor along the surface of the psoas muscle. We were able to gently lift the tumour and divide the right adrenal vein and arterial branches along the vena cava. Additionally, preoperative imaging studies are essential to map out the vascular anatomy and plan the surgical approach accordingly [13].

Last, but not least, the multidisciplinary team is essential in such complex oncologic cases. The collaboration among endocrinologists, oncologists, surgeons, radiologists, and pathologists is crucial for comprehensive diagnosis, treatment planning, and follow-up [2].

CONCLUSIONS

Laparoscopic adrenalectomy for large localized adrenocortical carcinoma remains a challenging, but feasible surgical option in carefully selected patients. This case report, along with the reviewed literature, demonstrates that with appropriate patient selection, meticulous surgical technique, and adherence to oncological principles, the complete resection of the tumour can be safely achieved.

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