# Images: Port site recurrence on followup imaging after adrenalectomy for adrenocortical carcinoma — first indicator of carcinomatosis

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## Introduction

Adrenocortical carcinoma (ACC) is a rare and aggressive malignancy of the adrenal cortex. Complete surgical resection is essential for localized tumours because ACC is highly resistant to chemotherapy and radiotherapy. Use of a laparoscopic approach for adrenalectomy in the setting of a confirmed or suspected ACC is controversial because it is unknown if laparoscopy provides equivalent oncological outcomes compared to an open approach.

# **Case report**

A 36-year-old male presented to his family physician following a workplace accident in which he sustained minor chest wall trauma. A series of investigations were performed to evaluate his injuries, including an abdominal ultrasound. The ultrasound incidentally detected a large right upper quadrant (RUQ) mass suspected to represent an adrenal malignancy. A referral was placed to the urology department at a tertiary care centre for further assessment. During the initial consultation, the patient reported significant weight loss (180 lbs) and mild RUQ pain. He denied facial flushing, headache, weakness, or polyuria. A computed tomography (CT) scan was arranged and revealed a 6 x 8 x 9 cm heterogeneous right adrenal mass with central necrosis (Fig. 1). These findings were consistent with a malignant lesion of the right adrenal gland.

Staging investigations, including an abdominal magnetic resonance imaging (MRI), chest CT, and bone scan, indicated the tumour was isolated to the adrenal gland, with no evidence of metastatic disease. Laparoscopic and open operative approaches were discussed with the patient and

after a thorough consultation, the decision was made to proceed laparoscopically given the patient's weight (124 kg) and the fact that imaging showing tumour isolated to the gland. The patient underwent a laparoscopic adrenalectomy within six weeks of his initial referral. The procedure was well-tolerated and he recovered appropriately. The surgical specimen is shown in Fig. 2.

Pathological review revealed a 10 cm, high-grade, adrenocortical carcinoma, stage pT2 (>5 cm, no extra-adrenal invasion) NX M0, as designated by the American Joint Committee on Cancer (AJCC). The neoplasm satisfied six of the nine Weiss criteria for the diagnosis of an adrenocortical malignancy, including diffuse architecture greater than one-third, clear cells ≤25% of total, significant nuclear pleomorphism, mitotic count ≥6 per 50 HPF, capsular invasion, and sinusoidal invasion. There was one small, focally positive margin. Medical and radiation oncology consultations were requested for consideration of adjuvant therapy. The patient was scheduled to receive six cycles of low-dose cisplatin (40 mg/m<sup>2</sup>) plus 50 Gray (Gy) of intensity-modulated radiotherapy (IMRT) in 25 fractions over a five-week period. He had significant side effects from chemotherapy, including nausea and thrombocytopenia, and was not able to complete the last of his six planned cycles. Adjuvant mitotane was also contemplated by medical oncology, but ultimately was not prescribed.

Thirty-three months after his adrenalectomy, the patient began to note progressive fatigue, weight gain, and depression. A CT scan was ordered and revealed four nodular lesions in the abdominal wall of his right flank (17 mm, 10 mm, 13 mm, 11 mm). The locations of these lesions were consistent with the laparoscopic port sites from his surgical resection (Fig. 3).

A multidisciplinary discussion was arranged and a decision was made to proceed with open resection of the four lesions. Intraoperatively, the patient was found to have multiple peritoneal deposits concerning for carcinomatosis in addition to the abdominal wall lesions detected on preoperative imaging (Fig. 4).

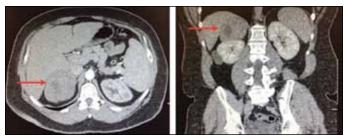


Fig. 1. Axial and coronal computed tomography images of heterogeneous right adrenal mass.

Frozen sections confirmed these deposits to be malignant, and all identifiable peritoneal lesions and abdominal wall masses were excised. The patient tolerated the procedure well and recovered without complication. Final pathological assessment confirmed all excised tissue was consistent with metastatic adrenocortical carcinoma. The patient is currently being evaluated for further systemic therapy with mitotane or inclusion in ongoing clinical trials by medical oncology.

### **Discussion**

ACC is a rare and aggressive malignancy of the adrenal cortex. Patients often present with advanced disease and have a poor overall prognosis. Complete surgical resection is essential, as ACC is resistant to chemotherapy and radiotherapy.<sup>1</sup>

This patient presented with recurrent disease that appeared to be isolated to the laparoscopic port sites nearly three years after his initial operation. Literature reviews published in 2008 reported 28 cases of port-site metastases following urologic oncology procedures.<sup>2-4</sup> The pathophysiological theory surrounding port-site recurrence is debated, but currently focuses on tumour seeding into the incisions at the time of surgery by direct tumour contact with the wound or surgical instruments that have been contaminated.<sup>4</sup>

There have been a number of studies that have evaluated the effectiveness of a laparoscopic adrenalectomy in the setting of a known or suspected ACC.<sup>5-9</sup> Multiple reports have raised concern regarding inferior oncological outcomes with a laparoscopic approach.<sup>5,8</sup> Other studies, however, have reported favourable outcomes, including masses >7

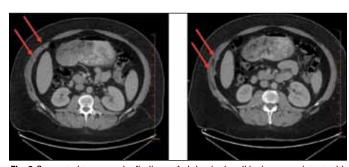


Fig. 3. Computed tomography findings of abdominal wall lesions consistent with laparoscopic port sites.



Fig. 2. Adrenocortical carcinoma resected by laparoscopic adrenalectomy.

cm, provided the lesion is confined to the gland.<sup>6</sup> A 2016 meta-analysis comparing open and laparoscopic adrenalectomy for ACC reported no difference in the overall recurrence rates, time to recurrence, or cancer-specific mortality;9 however, laparoscopic adrenalectomy was associated with a higher rate of peritoneal carcinomatosis. The authors concluded that open resection should be considered the standard surgical management of ACC, but a minimally invasive approach could be offered in carefully selected cases at centres with laparoscopic expertise. The results of this meta-analysis are congruent with current guidelines published by the Canadian Urology Association (CUA) and the Society of American Gastrointestinal and Endoscopic Surgeons (SAGES). Both guidelines discuss the necessity of complete resection in a specialized healthcare centre; however, each guideline also states that beginning a case with a laparoscopic approach is reasonable even if a malignant lesion is suspected. In our case, a laparoscopic approach was selected based on the patient's significant body mass index and absence of obvious invasion of other surrounding structures on preoperative imaging.

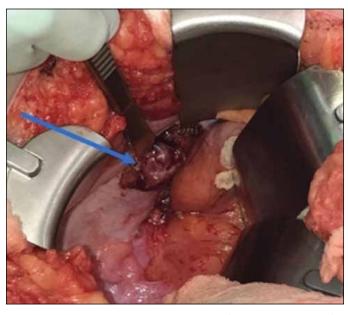


Fig.4. Intraoperative finding of abdominal wall mass (indicated by blue arrow).

# **Conclusion**

ACC is a challenging malignancy with high recurrence and mortality rates. Open adrenalectomy should be considered the standard of care when ACC is suspected and a minimally invasive approach should only be offered in carefully selected cases at centres with appropriate laparoscopic expertise. If a laparoscopic approach is selected, surgeons should have a low threshold for conversion to open adrenalectomy, as complete surgical resection is paramount for optimal oncological outcomes.

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Competing interests: Dr. Lavallée has participated in advisory board meetings for Ferring and Sanofi; and has received a grant from Sanofi. The remaining authors report no other competing personal or financial interests.

This paper has been peer-reviewed.

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