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 (Figure 1). These findings were consistent with a malignant lesion of the right adrenal gland.

Staging investigations including an abdominal 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 imaging showing tumour isolated to the gland. The patient underwent a laparoscopic adrenalectomy within 6 weeks of his initial referral. The procedure was well tolerated and he recovered appropriately. The surgical specimen is shown in Figure 2.
Pathologic review revealed a 10cm, high grade, adrenocortical carcinoma, stage pT2 (>5cm, no extra-adrenal invasion) NX M0, as designated by the American Joint Committee on Cancer (AJCC). The neoplasm satisfied 6 of the 9 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 6 cycles of low dose Cisplatin (40mg/m²) plus 50 Gray (Gy) of intensity-modulated radiotherapy (IMRT) in 25 fractions over a 5-week period. He had significant side effects from chemotherapy including nausea and thrombocytopenia and was not able to complete the last of his 6 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 (17mm, 10mm, 13mm, 11mm). The locations of these lesions were consistent with the laparoscopic port sites from his surgical resection (Figure 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. (Figure 4).

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
Adrenocortical carcinoma (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.1

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.2-4 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.4
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. Multiple reports have raised concern regarding inferior oncological outcomes with a laparoscopic approach. Other studies, however, have reported favourable outcomes, including masses >7cm, provided the lesion is confined to the gland. 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. 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 centers 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 health care 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 (BMI) and absence of obvious invasion of other surrounding structures on preoperative imaging.

Conclusion
Adrenocortical carcinoma 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 centers 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.
References

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

**Fig. 2.** Adrenocortical carcinoma resected by laparoscopic adrenalectomy.

**Fig. 3.** Computed tomography findings of abdominal wall lesions consistent with laparoscopic port sites.
Fig. 4. Intraoperative finding of abdominal wall mass (indicated by blue arrow).