CASE REPORT

Long-term recurrence-free survivor after laparoscopic removal of solitary adrenal metastasis from endometrial adenocarcinoma

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Abstract: Solitary adrenal metastasis from endometrial adenocarcinoma is extremely rare. We report herein the case of a laparoscopically resected solitary adrenal metastasis originating from endometrial adenocarcinoma. The patient was a 55-year-old woman who had undergone total abdominal hysterectomy for stage IIIc endometrial carcinoma, followed by 7 courses of adjuvant chemotherapy comprising carboplatin and paclitaxel. However, the patient developed an isolated right adrenal metastasis 15 months postoperatively. The solitary adrenal metastasis (diameter, 5.7 cm) was removed laparoscopically. The patient has now been in good health without recurrence for 5 years and 7 months after laparoscopic surgery. To the best of our knowledge, this is the first case of solitary adrenal metastasis originating from endometrial adenocarcinoma that is controlled for the long term by successful laparoscopic resection. J. Med. Invest. 57: 174-177, February, 2010

Keywords: laparoscopic adrenalectomy, solitary adrenal metastasis, endometrial adenocarcinoma

INTRODUCTION

The tumors that most commonly metastasize to the adrenal gland are lung cancer, breast cancer, gastric cancer, renal cell carcinoma, malignant melanoma, followed by liver cancer (1, 2). Patients may rarely present with solitary adrenal metastasis (2). In general, the presence of adrenal metastasis in endometrial adenocarcinoma indicates widespread disease (3). Solitary adrenal metastasis from endometrial adenocarcinoma is thus extremely rare. To the best of our knowledge, only two cases of isolated adrenal metastasis from endometrial adenocarcinoma have been reported (3), and this represents the first description of a laparoscopically resected solitary adrenal metastasis originating from endometrial adenocarcinoma.

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CASE REPORT

A 55-year-old woman presented in June 2004 with abnormal genital bleeding. She was referred to our hospital with endometrial adenocarcinoma in July 2004. Her serum carcinoembryonic antigen, CA19-9 and CA125 were within normal limits. The

patient underwent total abdominal hysterectomy with bilateral salpingo- oophorectomy and pelvic lymph node dissection. Histopathological examination revealed well-differentiated endometrial carcinoma with depth of myometraial invasion in the inner one-half, with no invasion of the cervix or adnexa. The cytological diagnosis of ascitis in the pelvic cavity was negative, but she had pelvic node involvement. We diagnosed stage IIIc. The patient then received 7 courses of adjuvant chemotherapy with carboplatin and paclitaxel. However, surveillance computed tomography (CT) performed 14 months postoperatively revealed a suspicious right adrenal tumor, 30 mm in diameter (Fig. 1), which



Fig. 1: Surveillance computed tomography performed 14 months postoperatively, revealing a right adrenal tumor, 30 mm in diameter

had not been present in previous scans. No other sites of metastatic disease were identified. CT performed 1 month later showed a rapidly enlarging adrenal tumor, 57 mm in diameter, indicating metastasis (Fig. 2). Laparoscopic adrenalectomy was performed. Mild adhesions existed between the tumor and liver or inferior vena cava, but the tumor could be successfully removed. Operative time was 3 h 50 min. Pathological examination was consistent with metastasis from the endometrial adenocarcinoma (Fig. 3). The patient was treated with 3 courses of adjuvant chemotherapy using carboplatin and paclitaxel after laparoscopic adrenalectomy. She has now been in good health without recurrence for 5 years and 7 months after laparoscopic surgery.

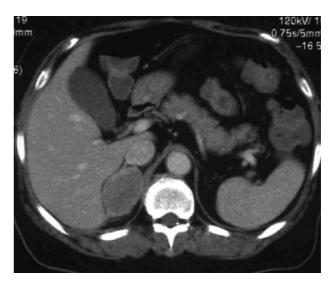


Fig. 2: CT showing rapid enlargement of the metastatic adrenal tumor, reaching 57 mm in diameter 15 months postoperatively.

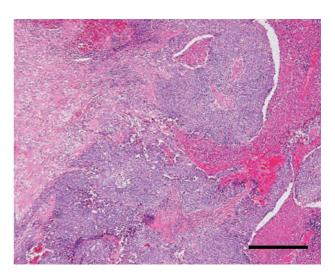


Fig. 3 : Histological appearance of metastatic adenocarcinoma (right) with compressed adrenal gland (left). The tumor displayed glandular and solid patterns with necrosis, compatible with endometrial adenocarcinoma. Bar, $40\,\mu m$.

DISCUSSION

Since the first report by Gagner et al. in 1991 (4), laparoscopic adrenalectomy has been adopted as the gold standard for surgical management of most adrenal tumors. Current indications for laparoscopic adrenalectomy include biochemically and clinically functional adrenal tumors and potentially malignant tumors of the adrenal gland (5). This indication applies to the open adrenalectomy. Potentially malignant tumors of the adrenal gland are generally considered to be non-functional tumors exceeding 5-6 cm in maximum diameter, including

adrenal metastasis or primary adrenocortical carcinoma. However, the role of laparoscopy in the treatment of adrenal malignancies remain controversial. In general, malignant tumor has the presence of dense adhesions, fixation of the tumor, and unusually numerous retroperitoneal feeding vessels. In this sense, the laparoscopic adrenalectomy for adrenal metastasis should be performed by expert laparoscopists (6). Laparoscopic adrenalectomy has been contraindicated in cases of proven or suspected primary adrenal cancer. The locoregional recurrence rate after laparoscopic adrenalectomy of primary adrenal cancer is 60%, occurring 1-2.5 years after resection. This recurrence rate resembles that for patients who have undergone open adrenalectomy for adrenocortical carcinoma (7). As for metastatic cancer, the presence of adrenal metastasis signals widespread disease. In general, the prognosis of patients with metastatic disease is poor, with few survivors at 5 years. In regard to non-small cell lung cancer, Luketich and Burt reported on 14 patients with isolated adrenal metastasis from lung cancer. Median survival was greater in the open surgical resection plus chemotherapy group of 6 patients (31 months) than in the chemotherapy alone group of 6 patients (8.5 months, p=0.03) (8). The treatment of isolated adrenal metastases mainly from primary lung cancer is advantageous to the patient if complete removal of the gland can be achieved (9, 10), whereas chemotherapy alone thus seems to have been largely ineffective against adrenal metastasis from non-small cell lung cancer. The same may apply to adrenal metastasis of endometrial adenocarcinoma. In general, the presence of adrenal metastasis from endometrial adenocarcinoma signals widespread or end-stage disease (3). Only 6% of patients with recurrent uterus cancer survived 3 or more years (11). A solitary adrenal metastasis from endometrial adenocarcinoma is extremely rare (Table 1). Combination therapy with carboplatin and paclitaxel reportedly covers the major effective drug

classes in the treatment of advanced and recurrent endometrial cancer (12-14). Overall response rates for patients with evaluable disease range from 62.0 to 82.1% (12-14). Our case was treated with 7 courses of adjuvant chemotherapy using carboplatin and paclitaxel after the initial operation, but surveillance CT performed 14 months postoperatively revealed the right adrenal tumor, 30 mm in diameter. CT 1 month later showed the adrenal tumor had grown to 57 mm in diameter. The volume-doubling time as calculated using the method originally described by Schwartz (15) was 13 days. We diagnosed this rapidly enlarging adrenal tumor as adrenal metastasis from endometrial adenocarcinoma. Chemotherapy thus seems to have been largely ineffective against adrenal metastasis from the endometrial adenocarcinoma. We therefore strongly recommend performing additional surveillance CT soon after detecting small adrenal tumor if the primary lesion was endometrial adenocarcinoma. In addition, surgery should be performed as soon as possible if the adrenal tumor is diagnosed as solitary metastasis of endometrial adenocarcinoma.

Diagnosis of the adrenal tumor as a solitary adrenal metastasis is very important. Several reports have described long-term survival after adrenalectomy for isolated adrenal metastases. The development of ¹⁸F-fluorodeoxyglucose (FDG) positron emission tomography (PET) as an imaging technique utilizing the fact that malignant cells contain increased levels of hexokinase (the enzyme responsible for glucose phosphorylation) appears promising for characterizing metastatic adrenal masses. The sensitivity, specificity, accuracy, positive predictive value, and negative predictive value of PET and/or PET/CT in detecting recurrences in patients with endometrial adenocarcinoma have been reported as 100, 83.3, 96, 95, and 100%, respectively (16). As we treated this patient before our hospital started running PET/CT, this modality was unavailable. Currently, all patients with confirmed or

| Table 1 : | Solitary adrena | l metastasis cases froi | m endometrial a | denocarcinoma |
|-----------|-----------------|-------------------------|-----------------|---------------|
|-----------|-----------------|-------------------------|-----------------|---------------|

| No | Age | Tumor size(mm) | Management of primary tumor | Management of adrenal tumor | status | Survival (months) |
|----|-----|-------------------|-----------------------------|-----------------------------|--------|----------------------|
| 1 | 76 | 50 | Laparotomy Radiation | Laparotomy chemotherapy | died | 15 |
| 2 | 61 | 50 | Laparotomy Radiation | Laparotomy chemotherapy | died | 12 |
| 3 | 55 | 57 | Laparotomy chemotherapy | Laparotomy chemotherapy | alive | 67 |

suspected solitary adrenal metastasis at our institution undergo PET with the aim of confirming the lesion as a solitary metastasis.

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