A Rare Case of Stage IV Adrenocortical Carcinoma with Acute Adrenocortical Dysfunction After Trans-Catheter Arterial Chemo-Embolization (TACE)

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Case report

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Abstract

**Background:** Adrenocortical carcinoma (ACC) is a rare and highly invasive endocrine malignant tumor with poor prognosis and insensitivity to chemotherapy, which don't have effective treatment. Although surgical resection is considered to be the main treatment for ACC, postoperative recurrence and metastasis have become the most important factors of death. Therefore, local treatments such as transcatheter arterial chemoembolization, radiofrequency ablation become be new treatment for ACC. Transcatheter arterial chemo-embolization therapy for ACC patients with liver metastasis has good efficacy and can effectively reduce the tumor burden of patients, which is considered to be safe and easy for patients.

**Case presentation:** We report a 47-year-old female patient diagnosed with stage II ACC with liver metastases, who developed symptoms of acute adrenocortical dysfunctions after hepatic arterial catheter chemo-embolization.

**Conclusion:** Trans-catheter arterial chemo-embolization therapy for ACC patients with liver metastasis is not completely safe, and there is a certain probability that it will lead to secondary adrenal dysfunction. Hydrocortisone supplementation can effectively alleviate the symptoms.

Background

ACC is a very rare tumor accounting for 0.7–2.0 cases/million people per year, occupying about 0.02% of all malignant tumors [1–2]. The treatment of ACC is still unclear in the world. Although surgical resection of tumor is considered to be the most effective method, the occurrence of tumor recurrence or distant metastasis still surrounds 80% of surgical patients [3]. In recent years, the treatment of recurrence and/or metastasis of ACC can be divided into three categories. The first category assesses ACC that can be completely removed by surgery, and surgical resection is possible. Due to the high risk of recurrence, oral mitotane after surgery is considered reasonable. The second category, such as inoperable ACC, local treatment is evaluated if there are indications for local treatment. The third category is ACC without local treatment conditions, you can choose mitotane alone or mitotane combined with streptozotocin palliative treatment according to the current research, and prefer the latter.

Adrenal dysfunction refers to a sharp drop in excessive cortisol levels, which leads to a series of adverse symptoms caused by adrenal corticosteroid deficiency. It often occurs after surgical resection of the adrenal glands, so glucocorticoids should be supplemented in time after surgery [4]. We report a 47-year-old woman diagnosed with stage II ACC with liver metastases, who developed acute adrenal insufficiency after hepatic arterial catheter chemo-embolization.

Case Presentation

A 47-year-old woman was admitted to our department after surgical resection for ACC. She was alert and conscious. According to the medical history, the patient underwent high-resolution computed
tomography (CT) of the chest, abdomen, and pelvis in May 2019, which indicated a large space occupying right adrenal gland with local necrosis (Fig. 1). The patient underwent right adrenal resection, and was defined as ACC in combination with immunohistochemical parameters and postoperative pathology [5]. Tumor cells were detected in the vena cava, and mitotane was taken all the time after surgery for ACC. On admission, the patient presented hypertension with blood pressure fluctuation of 169 – 139/101 – 90 mmHg, heart rate of 60–70 beats/min, no liver and kidney function impairment, and blood oxygen saturation of 98%, the hormone level: ACTH 36.4 pg/ml, cortisol 14.69 µg/dL. High-resolution computed tomography (CT) scans of the chest, abdomen and pelvis showed changes in adrenal resection, but the left lobe of the liver was a large circular mass, about 112.7*79.8 mm in size, with irregular density. This indicates that ACC recurrence is accompanied by liver metastasis, which is also one of the most common metastatic sites of ACC [6]. According to the Clinical Practice guidelines of the European Endocrine Society for the treatment of adult adrenal cortical carcinoma, local treatment may be beneficial for patients with advanced ACC with metastasis. We treated her with trans-catheter arterial chemo-embolization (TACE) for liver lesions. We choose to puncture the right femoral artery, and the 5F catheter sheath and the 5F RH catheter were inserted successively. The catheter head was inserted into the celiac artery for DSA imaging. A lightly stained large tumor was seen in the left lobe of the liver, and the left hepatic artery participated in the blood supply of the tumor. A microcatheter was used to superselect into the blood supply artery of the left hepatic artery tumor, and the mixed emulsion of 60 mg cisplatin + 10 ml iodized oil was injected, and an appropriate amount of 560 µm gelatin sponge particles were used to reinforce the embolism. The second imaging showed that the lipiodol deposition was acceptable and the blood supply artery of the tumor was blocked (Fig. 2). After the operation, anti-infection, liver protection, hemostasis, fluid infusion and other symptomatic treatment. In the first three days after the operation, the patient was accompanied by fever, mild liver damage, and blood pressure maintained at 104 – 100/70 – 60 mmHg. The treatment was treated with anti-infection, liver protection, and fluid infusion. On the 4th day, the patient’s fatigue, anorexia, fever, morning blood pressure decreased to 77/41 mmHg, heart rate 80 beats/min, hemoglobin 105 g/dL, K 2.56 mmol/L, Na 123.8 mmol/L, C-reactive protein 219.80 µg/L, fever to 39.2°C, We suspect that there may be Embolism syndrome. Hemostasis, dopamine raises blood pressure, electrolyte balance correction, and fluid infusion were treated. Afterwards, blood pressure remained at about 105/90 mmHg, but the patient’s condition has not improved significantly. Re-check the hormone level: ACTH 298.54 pg/ml, cortisol < 0.1 µg/dl. We believe that it is caused by decreased adrenal cortex function, but due to the possible risk of bleeding, we only give hydrocortisone 100 mg every other day and maintain dopamine boost. After that, the patient’s condition was stabilized, the blood pressure maintained at 136 – 120/95 – 82 mmHg, and the reexamination of CT showed that the lipiodol was deposited well and was discharged from the hospital (Fig. 3).

**Discussion**

ACC is a very rare malignant tumor, with an incidence of about 0.7-2.0 cases per 1 million people per year, accounting for about 0.02% of all malignant tumors [1–2], making clinical diagnosis become difficult. In
addition, 80% of ACC patients were found in the late stage of the disease [7], and lost the best time for surgery. However, after surgical resection, most patients will still have tumor recurrence or distant metastasis, leading to disease progression and death. At present, the 5-year overall survival rate of metastatic ACC is expected to be less than 15% [8]. For patients with metastatic ACC, according to the latest clinical practice guidelines of the European Endocrine Society for the treatment of adult adrenal cortical carcinoma [9]. Radiofrequency ablation, arterial chemoembolization and other methods to reduce the burden of local metastasis are commonly used clinically [10–11]. As previously reported, TACE is highly secure, but there are exceptions. The case of ACC we reported had recurrence with liver metastasis after surgery. We chose arterial catheter embolization to relieve liver load, but the patient continued to have fever, accompanied by hypokalemia and mild hyposodium in the first 3 days after treatment. By the 4th day, the patient had sustained blood pressure drop with fever. Although the patient had developed hypokalemia and low sodium, we did not notice the adrenal dysfunction, which was considered as post-embolism syndrome. Symptomatic treatments such as Dopamine raises blood pressure, anti-infection and rehydration were given to the patient, but the remission was not obvious. When we checked our hormone levels, we saw a sharp drop in cortisol and a sharp rise in adrenaline, and we realized that the patient had symptoms consistent with low blood pressure, fever, low potassium, and low sodium due to acute adrenocortical dysfunction.

Adrenal hypofunction often occurs after adrenal gland removal or long-term use of glucocorticoids, etc [12–13]. As far as we know, adrenal hypofunction is a very rare phenomenon after arterial duct embolization, and it has not been reported so far. These complications are extremely dangerous if the patient is discharged from hospital because of a doctor’s error of judgment. Acute adrenocortical dysfunction can be found in early time, such as timely reexamination of ACTH, cortisol and other hormones, timely supplementation of glucocorticoid to avoid death, so early diagnosis is particularly important. We reported this case, mainly to remind that such symptoms occurred after catheter embolization chemotherapy, and should be associated with acute adrenal hypofunction caused by treatment as early as possible. And we also believe that catheter embolization has a good chemical effect, leading to a large number of tumor cell deaths and a sharp decrease in the secretion of cortisol, leading to a sharp decline in acute adrenal cortex function.

**Conclusion**

We have experienced a rare case of acute adrenal hypofunction caused by ACC trans-arterial catheter embolization chemotherapy. The symptoms of this case were relieved by supplementing hydrocortisone, which also makes us realize the importance of reexamination of hormone levels in ACC patients after TACE.

**Declarations**

**Ethics approval and consent to participate**
The study plan was approved by the ethics committee of the First Affiliated Hospital of Anhui Medical Science University, China.

Consent

The case report, including relevant images, has been published and the patient’s written informed consent has been obtained.

Availability of data and materials

Data sets and images used in this study may be provided by corresponding authors upon reasonable request.

Competing interests

All authors declare that they have no competing interests.

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None.

Authors’ contributions

LP helps to study the concept. SX, ZFL, and WT contribute significantly to case studies, manuscript preparation, and discussion. WZL conducted case analysis, manuscript preparation and discussion, and wrote the manuscript. All authors approve the final version for publication.

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