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Case Report

Multiple brain metastases in a patient with uterine papillary serous adenocarcinoma: Treatment options for this rarely seen metastatic brain tumor

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Abstract

Background: Uterine papillary serous adenocarcinoma (UPSAC) occurs 10-fold less frequently than endometrial carcinoma, and is referred to type 2 endometrial adenocarcinoma. The prognosis of UPSAC is worse than that of type I endometrial carcinoma. Herein we report what is only the second case of UPSAC, but it should prove to be more informative than the first reported case.

Case Description: A 71-year-old female had three different metastases in the brain; two of the metastases were located in the posterior fossa within the cerebellar parenchyma with perilesional edema, but no mass effect, and the third metastasis was located in the right frontal lobe, and caused hemispheric edema and subfalcine herniation. The lesion that caused mass effect was completely extirpated without any surgical complications. The patient's recovery was excellent. She is able to walk independently, and use her left hand and left arm. Her Karnofsky performance score 5 months postsurgery was 80/100.

Conclusion: Based on the outcome in the presented case, we think that in any UPSAC patient with a metastatic brain tumor causing mass effect the symptomatic metastatic tumor must be removed, regardless of disease grade, to ensure optimal quality of life.

Key Words: Adenocarcinoma cerebrum, cerebellum, metastases, uterine

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INTRODUCTION

Hendrickson *et al*. were the first to describe uterine papillary serous adenocarcinoma (UPSAC) (also known as type 2 endometrial carcinoma), an aggressive form of endometrial carcinoma.[8] Although UPSAC accounts for <10% of all endometrial carcinomas, little is known about the clinical course of this type of uterine malignancy because of its rarity.[1,2,7,9] Patients with UPSAC are classified into four groups based on human epidermal growth factor 2 (HER2) and hormone receptor expression (HR).[1,2,7,9] HER2 overexpression was correlated with a lower overall survival rate, but HR overexpression was correlated with a higher survival rate. The 5-year recurrence-free survival rate to overall survival rate ratio in this group is 43%:51%.[1,4,6,14,15]

CASE REPORT

A 71-year-old female underwent abdominal hysterectomy with bilateral salpingo-oopherectomy 2 years and

3 months ago. Histopathological examination of the biopsy specimen showed UPSAC. Adjuvant chemotherapy was given after the abdominal hysterectomy. The patient's Glasgow Coma Scale score was 12/15 at the time of admission to our hospital. She presented with a 20-day history of headache and left hemiparesis, and she could not walk or use her left arm and left hand. Cranial magnetic resonance iaging (MRI) showed three lesions; two were located in the parenchyma of the cerebellum and had perilesional edema. but had no shift effect or obstruction on the fourth ventricle and the third lesion was located in the right frontobasal frontal lobe, and caused a large hemispheric edema and subfalcine herniation [Figure 1a-d]. All the three lesions exhibited contrast enhancement following gadolinium injection. The lesions' characteristics are shown in Figures 1-3.

The patient underwent frontal craniotomy with gross total extirpation of the tumor located in the right frontal lobe on September 12, 2012. There were no surgical complications, such as bleeding or adhesiveness adjacent to the brain tissue during the removal of the tumor. The other two lesions located in the posterior fossa were left untouched because they had no mass effect. Histopathological examination of the biopsy specimen was consistent with metastatic UPSAC [Figure 4].

The patient was able to walk with the assistance of a walker, and underwent follow-up cranial computed tomography (CT) 2 days postsurgery [Figure 5], which showed that there was no residual tumor in the frontal lobe, the midline shift and subfalcine herniation improved, and less edema than presurgery. The patient was followed-up for 9 months postsurgery and via telephone (last contact on May 14, 2013) reported that she was able to walk independently, and use her left arm and left hand without any difficulty, and that she had undergone 10 days whole cranial radiotherapy and chemotherapy at another hospital. The contrast enhanced cranial MRI was performed following these therapies on February 18, 2013. It showed no residual or recurrent lesion at the operation site, and no lesion at the cerebellum, which had had two lesions in previous MRI. In addition, the edema was remarkably lessened at the right cerebral hemisphere regarding preoperative and early postoperative period.

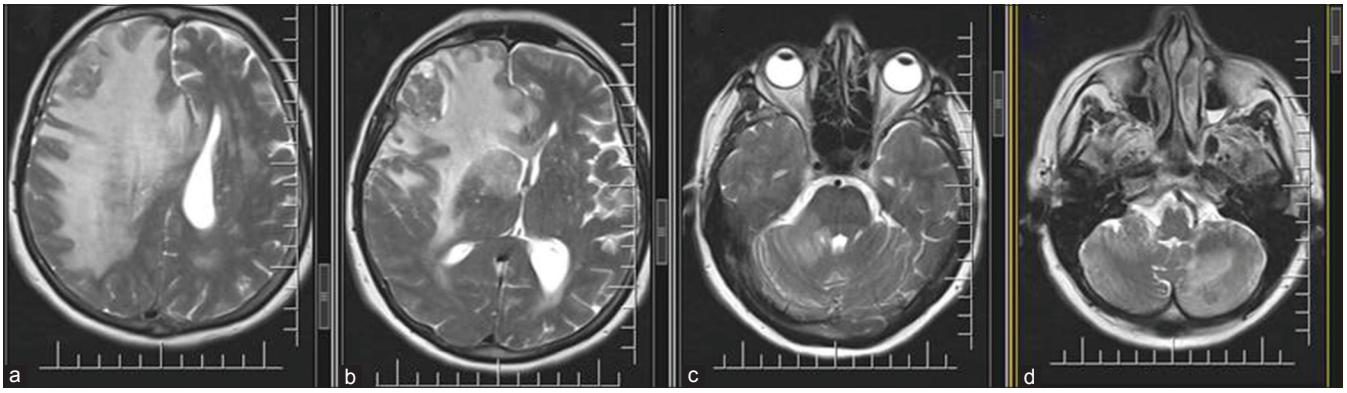

Figure 1: T2W1: (a, b axial supratentorial; c, d, axial infratentorial) (a and b): MRI show massive edema on the right frontoparietal region and midline shift. (c and d): MRI show minimal edema on the right and left cerebellum and minimal mass effect

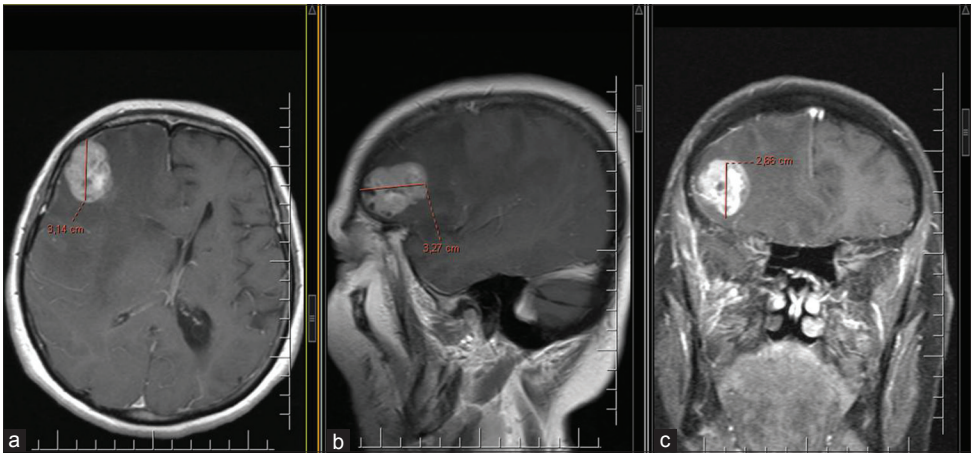

Figure 2: (T1W1: a, axial; b, sagittal; c, coronal.) Postcontrast MRI show enhancing lesion which is well defined and relatively homogenous and is located in the base of the right frontal lobe, and it is just above the orbital roof. The mass causes edema, midline shift and subfalcine herniation due to massive edema in the right cerebral hemisphere. The size of the lesion is 33 × 32 × 24 mm in three different sections

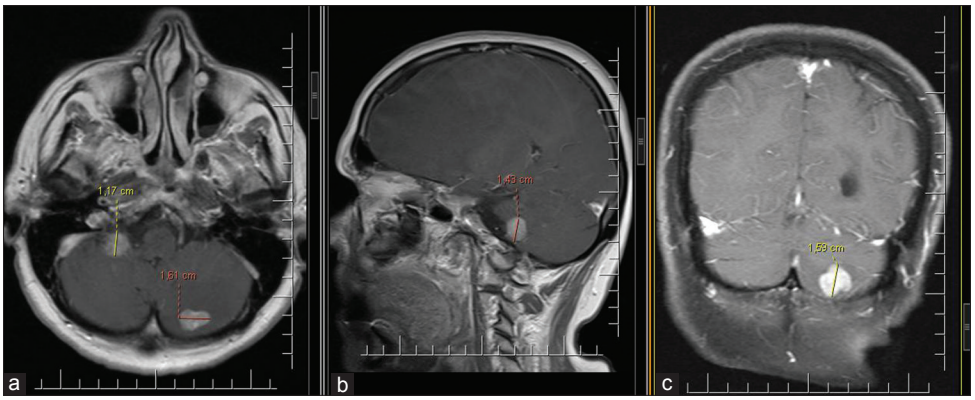

Figure 3: T1W1: (a, axial; b, sagittal; c, coronal.) Postcontrast enhanced MRI show enhancing two different mass lesions in the cerebellum. Both of these lesions cause minimal mass effect. The size of the lesion which is placed around right inferior cerebellar peduncle is 11 × 15 × 12 mm in three different sections

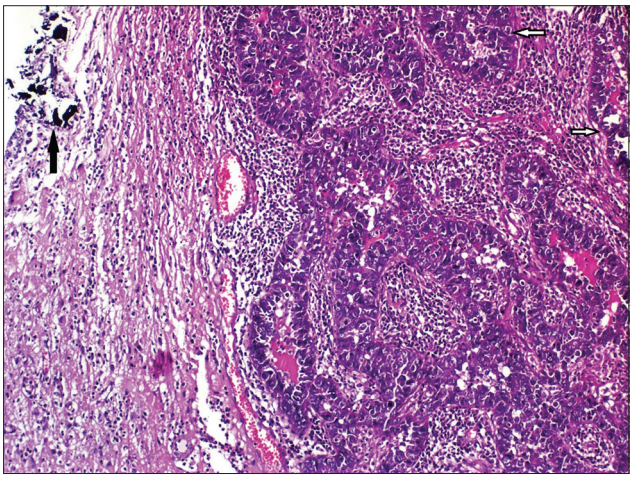

Figure 4: Brain metastatic endometrial serous type carcinoma. A nodule of metastatic neoplastic glands with luminal micropapillary structures (white arrows) compresses adjacent glial tissue (left side) showing marked reactive gliosis, lymphoplasmositer infiltrate and microcalcifications (black arrow). Hematoxylin and eosin (H and E) stain ×100

DISCUSSION

Metastatic brain tumors are the most common brain tumor and have an annual incidence rate 10-fold higher than primary brain tumors.[10,17] Metastatic brain tumors in adults originate primarily from the lungs, breasts, kidneys (renal cell), and gastrointestinal system.[10,17] Gynecologic malignancies metastasize via implantation rather than hematologically, and therefore metastasize in the brain less frequently than in the lungs and other organs.[10,12,17] Metastatic brain lesions occur most frequently at the junction of the frontal, temporal, parietal, and occipital lobes, due to embolic spread to the terminal braches of the middle cerebral artery.[12] In addition, they tend to arise at the gray matter–white matter interface. Invasion to the cerebellum can occur via the spinal epidural venous plexus (Batson's plexus) and the vertebral veins. Moreover, the uterus has a very close anatomical relationship with Batson's plexus via the sacral plexus. Lastly, venous blood from Batson's plexus reaches the pulmonary vasculature and dissemination ensues via the pulmonary vasculature.[11,12]

The posterior fossa–or cerebellar parenchyma–is the second most common location of metastatic lesions, accounting for 16% of all cranial metastases.[12] UPSAC–an aggressive subtype of endometrial cancer also known as type II endometrial carcinoma–is uncommon and is associated with a poor prognosis. For example, the 5-year overall survival rate in patients with stage I UPSAC is 45-78%.[4,6,14,15] As UPSAC implants in the intraperitoneal region early, it is more aggressive and is associated with a worse prognosis than type I uterine carcinomas. HER2 positivity and HR negativity are responsible for the aggressiveness of UPSAC.[4,6,14,15] Cerebral or cerebellar metastasis of UPSAC is rare, and a search of the literature showed just one reported case of UPSAC with cerebellar metastasis, but the report did not provide a specific explanation of its pathology or brain MRI findings.[5] Furthermore, to the best of our knowledge the literature does not contain any report of multiple UPSAC metastases or surgical treatment for a metastatic tumor of UPSAC origin. In addition, the metastatic lesion located in the posterior fossa in the previously reported case did not cause any shift or edema, as in the presented case, but the supratentorial metastatic lesion located at the frontal lobe in the presented case caused hemispheric edema. As such, it can be said that a metastatic lesion of UPSAC origin located in the cerebellum may cause less edema, but a lesion located in the frontal lobe can cause excessive edema, which may lead to development of subfalcine herniation.

Metastasis of UPSAC to the cerebrum in the presented case was histopathologically confirmed, and to the best of our knowledge the presented case is the first to undergo surgical resection due to metastatic UPSAC in the cerebrum. The presented case was free of any brain metastasis 27 months following hysterectomy, which

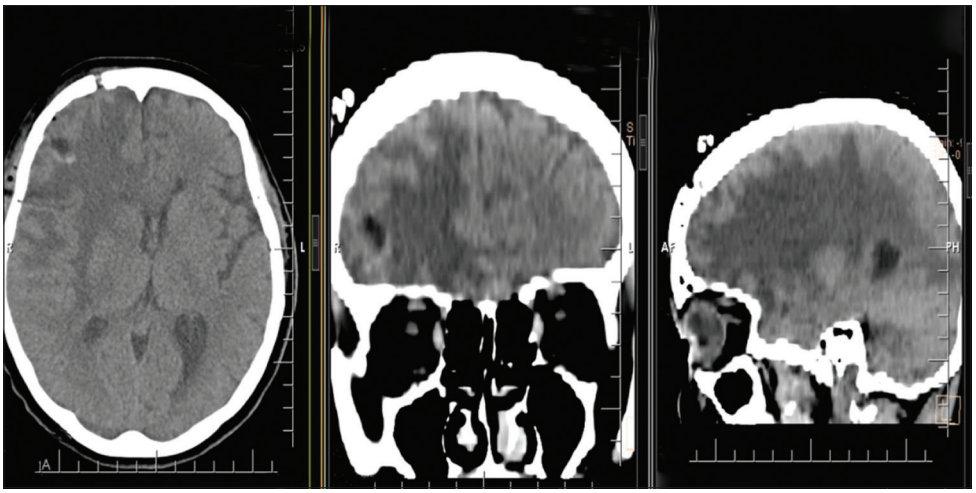

Figure 5: Axial, coronal, sagittal CT without contrast shows less edema and less midline shift than preoperative period. Besides, it shows total resection of the lesion located at the right frontal lobe

is consistent with the literature regarding endometrial carcinoma, but we did not find any data concerning brain metastasis with UPSAC in the literature.[3,13,16] We think that metastatic lesions of UPSAC origin located in the cerebrum can cause edema that can lead to development of subfalcine herniation or transtentorial herniation, according to the location of the metastatic lesion within the cerebrum. As such, we recommend removing such metastatic brain lesions that cause symptoms in patients with UPSAC, but only if the patient's general health status is sufficient. It is not possible to predict the effect of such surgery on the overall survival rate in such patients because of the systemic effect of HR negativity and HER2 positivity, which strongly influence survival.[4] Nevertheless, one should not hesitate to remove symptomatic metastatic lesions, so as to improve patient quality of life. In addition, chemotherapy and radiotherapy should be scheduled immediately postsurgery with a radiation oncologist and medical oncologist, because the response of these treatments would provide better life quality.

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