

## CASE REPORT

# Intracerebral hemorrhage cause by a ruptured oncotic aneurysm from choriocarcinoma metastasis

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## ABSTRACT

Ruptured oncotic aneurysms from choriocarcinoma metastasis are very rare. One rare case of metastatic cerebral choriocarcinoma with an initial presentation of intracerebral hemorrhage is reported. A 25-year-old woman initially presented with sudden onset of alteration of consciousness. The computed tomography scan showed left parietal hematoma and CT angiography showed a small aneurysm of the left middle cerebral arteries territories. Her chest X-ray showed two lung masses. A craniectomy was performed to remove the hematoma and decompression following the aneurysm excision. After surgery the patient regained consciousness and had no neurological deficit. The pathology reported metastatic choriocarcinoma and her beta – HCG was 73,656 units. After recovery she received chemotherapy with an etoposide, methotrexate, actinomycin D, cyclophosphamide, and vincristine/ovcovine (EMA-CO) regimen.

**Key words:** Cerebral metastases, choriocarcinoma, oncotic aneurysm

## Introduction

Choriocarcinoma is a rare, highly malignant neoplasm of trophoblastic origin in gestational trophoblastic disease. This tumor is known for its association with molar pregnancy, rapid hematogenous spread to multiple organs, high human chorionic gonadotropin (hCG) levels and good response to chemotherapy.<sup>[1]</sup> It is known that about 80% of gestational choriocarcinoma have remote metastasis lesions and the incidence of brain metastasis is the second highest next to lung metastasis. Cerebral metastasis occurs in 10% to 20% of patients with choriocarcinoma.<sup>[2]</sup> Many patients with brain metastasis develop sudden neurological involvement due to intracranial hemorrhage. The suspected cause of hemorrhage is bleeding from aneurysms that develop due to neoplastic cell invasion of the arterial wall.<sup>[3,4]</sup>

## Case Report

A 25-year-old woman presented with sudden onset of loss of

consciousness for two hours. No history of hypertension or bleeding disorders could be identified. Non enhanced head computed tomography (CT) was performed revealing a large subcortical hematoma in the left parietal area [Figure 1]. The CT angiography (CTA) revealed a small aneurysm in the periphery of the middle cerebral artery (MCA) [Figures 2-4]. The Glasgow Coma Score before surgery was seven. An emergency craniotomy was performed to remove the hematoma. A pseudo aneurysm of 0.8 cm. in diameter with perforation was identified at the left parietal lobe. The aneurysm was excised smoothly and a craniectomy was done secondary to brain swelling. The patient regained consciousness postoperatively with no neurologic deficit. Immunohistochemical staining of the aneurysm revealed a positive cytokeratin and beta – hCG [Figures 5-7]. A chest X-ray showed multiple lung masses giving the impression of lung metastasis. Based on these studies, the diagnosis of metastatic choriocarcinoma was made. Serum beta – hCG was 73,656 ng/ml. After diagnosis, the patient received chemotherapy with EMA-CO regimen. Her beta – hCG decreased from 73,656 ng/ml to 3.8 ng/ml after three courses of chemotherapy.

## Discussion

Choriocarcinoma has a marked tendency to be metastasized by blood borne dissemination. It is known that about 80% of choriocarcinomas have remote metastatic lesions. Choriocarcinoma has a tendency to rapidly and hematogenously spread to the lung, vagina, brain, liver, kidneys, and ovaries. Favored sites of involvement are the lungs (94%) and vagina (44%), followed by the liver (28%), and the brain (28%).<sup>[4,5]</sup> Metastatic choriocarcinoma involves the brain in 3 to 28% of

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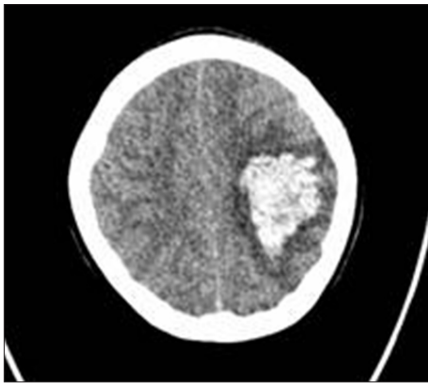


Figure 1: CT scan showed subcortical hematoma in left parietal area

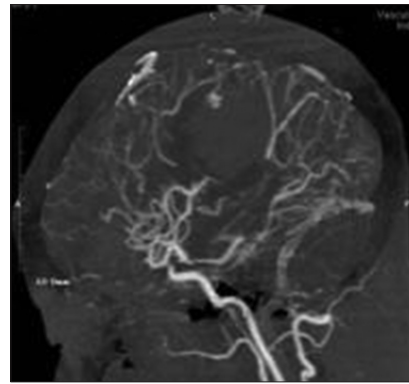


Figure 2: CTA showed small intracranial aneurysm above hematoma

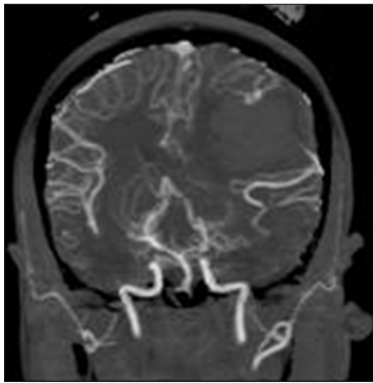


Figure 3 : CTA showed small intracranial aneurysm above hematoma

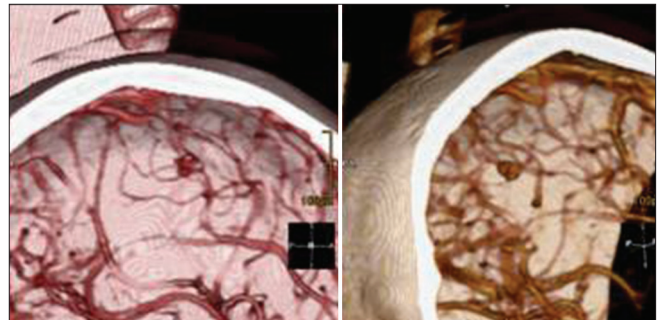


Figure 4: CTA showed small intracranial aneurysm at MCA territories branch

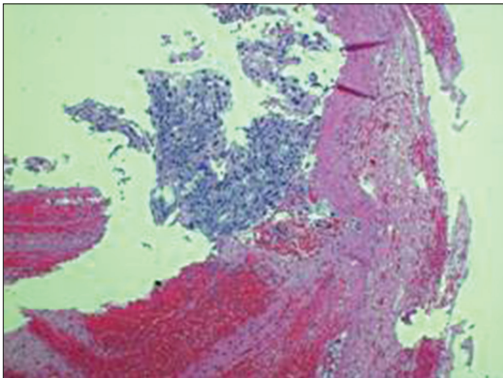


Figure 5: H and E stain showing tumor cells in aneurysm wall

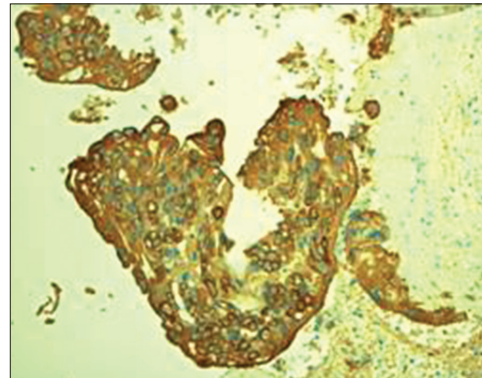


Figure 6: Cyokeratin stain in tumor cells

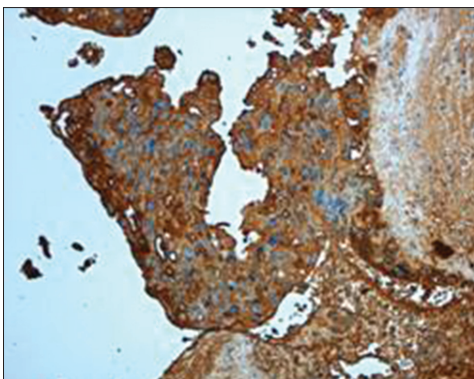


Figure 7: HCG stain in tumor cells

patients. Metastases are seen in decreasing order of frequency from the parietal to the temporal to the frontal lobes. Cerebral involvement is generally seen in patients with advanced disease. All patients with brain metastasis have concurrent pulmonary or vaginal involvement or both. Brain metastases are thought to arise from tumor emboli from the lungs, as it is most uncommon for such lesions to arise without pulmonary metastasis.<sup>[2]</sup>

Because of the innate capacity of trophoblastic cells to invade and erode the vessel wall, many patients with brain metastasis develop sudden neurological involvement due to intracranial hemorrhage. The intracranial hemorrhage may be subdural,

subarachnoid, or intracerebral. The most common symptoms are headache, nausea, vomiting, hemiparesis, depressed level of consciousness, and seizure.<sup>[1]</sup>

Single or multiple aneurysms are sometimes seen in choriocarcinoma. However, there are comparatively few cases in which the presence of aneurysms have been proven angiographically or histologically.<sup>[6,7]</sup> Less than 20 cases of ruptured oncotic aneurysms were reported in the literature, and most of them were located on branches of the middle cerebral artery. The angiography usually reveal a single, peripheral, and irregular aneurysm, but proximal and multiple aneurysms were also described.<sup>[8,9]</sup> The neoplastic aneurysms are postulated to occur because partial destruction of the vessels wall occurring after a necrotic embolus lodges in the vessels. Histological examination of the aneurysms showed occlusion of the vessels by tumor cell emboli, proliferation of the tumor cells into the vessels walls and rupture of the internal elastic lamina.<sup>[10,11]</sup>

Metastatic choriocarcinoma must be treated as soon as possible because this tumor has a tendency to grow rapidly and spread hematogenously. Surgery is associated with a high incidence of hemorrhagic complications and should only be performed if the hemorrhage is life threatening. A craniotomy is required to provide acute decompression or to control bleeding if cerebral lesions induce hemorrhage.<sup>[6]</sup> Moreover, there are reports in the literature of complete resolution of cerebral metastasis, oncotic carotid - cavernous sinus fistulas and oncotic aneurysms with chemotherapy.<sup>[6,8,12]</sup>

Chemotherapy is indicated in virtually all patients with choriocarcinoma, and it is appropriate that the regimen of chemotherapy be selected depending on the expected prognosis. It is well known that the prognosis of choriocarcinoma is affected by the existence of metastasis, patient's age, duration of chemotherapy, hCG levels, size of the tumor, and preceding pregnancy. More intensive chemotherapy is indicated for patients with remote metastases. Recently, combination therapy with etoposide, methotrexate, actinomycin-D, cyclophosphamide, and vincristine (EMA-CO) has been considered the most effective regimen for patient in the high risk group.<sup>[13,14]</sup> Patients who developed resistance or recurrence after EMA-CO regimen may then successfully be treated with a modification of the regimen by substituting etoposide and cisplatin on day 8 with the EMAEP regimen.<sup>[1]</sup>

## Conclusion

In conclusion, this case illustrates many points; first, the importance of biopsy in vascular lesions of undetermined etiology; second, angiographic studies are recommended in young patients with metastatic choriocarcinoma who present with intracerebral hemorrhage.

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