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## CASE REPORT



# Othello Syndrome in a Patient with Two Left Hemispheric Tumors

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We report a case of a patient with Othello syndrome caused by two left hemispheric tumors. This 50-year-old female had experienced seizures for 10 years and developed manic-like symptoms, delusions of jealousy, persecution and being watched, auditory hallucinations, irritable mood, and violent and disorganized behavior for the past 3 years. Brain imaging studies revealed two left frontal tumors, the larger of which was causing a mass effect. The delusions of jealousy in Othello syndrome resolved after removing the larger tumor, and the other psychiatric symptoms improved after treatment with psychotropic medications. This report aims to raise awareness of Othello syndrome related to disruptions in cortico-subcortical connections in the left orbitofrontal region. Timely surgical treatment may prevent associated psychiatric comorbidities and increase the likelihood of a good outcome.

Key words: Delusions of jealousy, left hemispheric tumors, Othello syndrome

### INTRODUCTION

Othello syndrome is a type of morbid jealousy caused by a variety of factors, and it is associated with a variety of etiologies. Neurological diseases are the major cause of Othello syndrome<sup>1</sup> including epilepsy, cerebral infarction or hemorrhage, dementia, Parkinson's disease, encephalitis, normal pressure hydrocephalus, multiple sclerosis, delirium, and tumors.<sup>1-3</sup> Herein, we present a patient with two left hemispheric meningiomas causing Othello syndrome which resolved after removing one of the tumors and treatment medication of aripiprazole 20 mg/day for 3 months. The findings of this case may implicate a relationship between specific areas of the brain and Othello syndrome.

#### **CASE REPORT**

A 50-year-old homemaker presented with a history of epilepsy for which she had been taking phenobarbital 210 mg/day and vigabatrin 1000 mg/day for about 10 years. She had been experiencing episodic manic-like symptoms

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including an irritable mood, euphoria, and increased goal-directed behavior for the past 3 years, in waxing and waning course. She also suffered from jealous delusions, in which she accused her husband of having an extramarital affair, with one of his business partners for the last 2 years without evidence. She subsequently developed secondary delusions to those of jealousy, including being watched and persecuted, auditory hallucinations, an irritable mood, and violent and disorganized behavior. She was found wandering the streets on several occasions, which may have been associated with the jealous and persecutory delusions. She only took antiepileptics as aforementioned with antipsychotics at that time, even though her delusion of jealousy persisted. On December 12, 2014, she was sent to our emergency department because of generalized tonic-clonic seizures which had occurred twice that day. She also presented with headache, dizziness, nausea, and vomiting, and her family also reported a recent personality change. She did not exhibit any external injuries, neck

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stiffness, or other physical abnormalities. In addition, none of the laboratory examinations demonstrated any abnormal findings suggesting a medical disorder, and her urine toxicology screen was negative. Computed tomography of her brain showed two high-density masses in her left frontal lobe. She was then transferred to our neurosurgery Intensive Care Unit, given antiepileptic medications, and prepared for surgery. T1- and T2-weighted magnetic resonance imaging revealed two heterogeneous enhanced tumors, the larger of which was  $4.4 \text{ cm} \times 4.5 \text{ cm} \times 4.3 \text{ cm}$  in size in the left frontal region with perifocal edema compressing the frontal horn of bilateral ventricles with a midline shift to the right side. The smaller of the tumors was also located in the left frontal region near the central sulcus and was about  $4.0 \text{ cm} \times 2.1 \text{ cm}$ in size. Partial enhancement was noted with a lesser mass effect than with the large tumor [Figure 1]. She underwent surgical treatment with resection of the larger tumor, which was pathologically proven to be Grade I meningioma. The other tumor was not removed due to the lesser mass effect and limited surgical window with which to approach it. Her jealous delusions ceased after surgery. However, her persecutory delusions and behavioral disturbances persisted, so she was referred to our psychiatrist. She was prescribed with aripiprazole 20 mg/day, and she was in a relatively stable condition 3 months later. During 6 months of follow-up, she had no recurrence of the psychotic symptoms, and aripiprazole was gradually tapered off.

### DISCUSSION

The frontal lobe is associated with higher mental functions such as motivation, planning, social behavior, and speech production. Othello syndrome occurs most frequently with

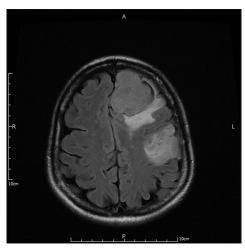


Figure 1: Magnetic resonance image of a 50-year-old female with Othello syndrome

neurological disorders, and the delusions appear to be associated with dysfunction of the frontal lobes, especially in the right frontal lobe where lesions have been reported more frequently than in the left.<sup>1,4</sup> Othello syndrome with right cerebral vascular infarction has been reported to be associated with the interruption of complex connections between the right hemisphere and frontal lobe.<sup>5</sup>

The orbitofrontal cortex of the frontal lobe mediates response inhibition, impulse control, social behavior, emotional regulation and generalized arousal through limbic nuclei, the dorsal medial nuclei of the thalamus, and reticular formation.<sup>6</sup> Othello syndrome secondary to right orbitofrontal lobe excision and right orbitofrontal lobe cavernous sinus hemorrhage has been reported.<sup>7,8</sup> In our reported case, her morbid jealousy improved significantly after excision of the extra-axial tumor in the orbitofrontal region of the left frontal lobe, while her other psychiatric symptoms including auditory hallucinations and delusions of persecution and being watched improved with antipsychotic treatment. However, Othello syndrome has also been reported to be associated with the coexistence of thalamic and left frontal lobe lesions. No lateralized brain dysfunctions have been reported to be associated with Othello syndrome.9 To the best of our knowledge, this is the first report of two tumors in the left hemisphere of the brain causing Othello syndrome. The jealous delusions of our patient improved after excision of the extra-axial tumor in the orbitofrontal region of the left frontal lobe, and her other psychiatric symptoms including the auditory hallucinations and delusions of persecution and being watched improved with antipsychotic treatment. The resolution of direct tumor compression and mass effect by surgical removal may have contributed to these improvements in her symptoms related to Othello syndrome, 10 which suggests that disruptions in cortico-subcortical connections might be one of the important contributing factors to the pathogenesis of Othello syndrome in the left frontal lobes. The resection of the tumor in the left frontal region with perifocal edema compressing the frontal horn of bilateral ventricles resulted in the resolution of jealous delusion, but the persecutory delusions and other behavioral disturbances resolved after 3-month of aripiprazole treatment as 20 mg/day. Therefore, the direct compression and mass effects of different tumor sites could be important in contributing in the different types of delusions. However, further study is needed to clarify this "brain-behavior" relationship. We hope that this report raises awareness of Othello syndrome in the tumors of left frontal lobes. The early recognition of the association between the psychotic symptoms and left hemispheric, frontal lobe tumors is important, and the timely antipsychotic treatment and surgical intervention may prevent associated psychiatric comorbidities and improve the likelihood of a good outcome.

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#### **Conflicts of interest**

There are no conflicts of interest.

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