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Secondary Mania After Pontin Cavernous Angioma

SIR: Secondary mania has been reported with toxic and metabolic disturbances, primary and metastatic brain tumors, epilepsy, and cerebrovascular events. We present a patient who developed a manic episode after a pontin cavernous angioma. To our knowledge, this is the first report describing secondary mania in a case with vascular angioma in the pons.

Case Report

A 34-year-old man was admitted to our department with the chief complaint of significant personality changes. His family reported enhanced talkativeness, increased psychomotor activities, increased sex drive, and decreased need for sleep for 1 week. He was unable to perform his job and his social activities were impaired. During a mental status examination, he was alert but his distractibility was outstanding and he did not establish eye contact. There were no perceptional abnormalities. The patient had rapid, increased talking and laughing and had grandiose ideas. He was overly excited and euphoric. Abstract thinking, judgment abilities, and insight were not impaired. His mood was anxious and he became irritable when hindered. The patient frequently acted in an impulsive way. He had no premorbid psychiatric and medical history, family history of affective disorder, or alcohol or substance addiction. His neurological examination was intact.

One day after his admittance, the patient was examined with a structured psychiatric interview and scales for measurement of impairment in activities of daily living, intellectual function, and social functioning. His Young Mania Rating Scale (YMRS) score was 46. Routine laboratory tests including thyroid hormones and thyroid-stimulating hormones were within normal limits. T2-weighted magnetic resonance images showed a well-circumscribed, lobulated lesion with a heterogenous core due to varying stages of blood products and surrounded by a rim in pons. These findings indicated cavernous angioma. Digital subtraction angiography was considered normal.

We administred a regimen of carbamezapin, 400 mg/d, but the symptoms continued. Two weeks later, he developed jerk nystagmus in primary position, vertical and lateral gaze, postural tremor, truncal ataxia, and left hemiparesis while his manic symptoms lasted. Two months later, a midline suboccipital craniectomy was performed and the lesion was dissected. Following surgery, psychomotor activation decreased, speech slowed down and sleep was regulated. We discontinued drugs. One month later the level of YMRS decreased to 20 points. His manic symptoms improved but nystagmus and slight left hemiparesis persisted.

Comment

10% to 30% of intracranial cavernomas are located in the posterior cranial fossa.[1] Characteristic symptoms of brainstem cavernous malformations include headache, emesis, ataxia, nystagmus, diplopia, hemiparesis, sensory changes, and change in mental status.[2] Several studies demonstrated a significant association between secondary mania and lesions involving cortical and subcortical regions of the right hemisphere. In mania, there is an increase in dopamine, serotonin, and noradrenaline transmission from the substantia nigra, raphe nuclei, and locus coeruleus to all areas of the brain, compared with a nondiseased brain. It seems that the main brain areas involved in mania include the frontal and temporal lobes of the forebrain, the prefrontal cortex, the basal ganglia, and parts of the limbic system. However, a dysfunction in any brain region associated with these mood-regulating circuits may lead to the development of a mood disorder.[3,4] Drake et al. reported two cases of people who developed mania after ventral pontine infarction and suggested that brainstem disturbances can influence mood, sleep, libido, and thought.[5] In reviewing the literature, we did not find that mania occurs because of any vascular malformation in the pons. It is possible that abnormalities in these circuits confer a biological vulnerability, which, when combined with environmental factors, causes mood disorders.

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Pathological Hyperfamiliarity for Others From a Left Anterior Cingulate Lesion

SIR: Person misidentification syndromes usually involve the misperception of familiar persons or a loss of familiarity for normally perceived ones. We report a unique patient who developed the opposite, a sense of hyperfamiliarity for unfamiliar persons after a hemorrhage in the left anterior cingulate cortex (ACC).

Case Report

A 34-year-old man developed an acute onset of a severe headache, followed by transient loss of alertness. He spontaneously recovered but 2 days later was hospitalized because of new-onset generalized seizures. The patient had a negative medical history and was on no medications at the time of admission.

On initial examination, he was in a postictal delirium. He had otherwise normal examinations of his cranial nerves, motor systems, and reflexes. He was loaded with phenytoin without further seizures. A computed tomography (CT) scan revealed a hyperdense region located in the left anterior cingulate cortex consistent with a hemorrhage. Angiography and magnetic resonance imaging (MRI) revealed a cavernous angioma. On electroencephalography, there were theta waves in the left frontocentral region.

One day after admission, mental status examination revealed an akinetic mute state. He was totally indifferent to external stimuli. Over the following days, the patient had gradually increasing verbal output and behavioral initiation. On day 7 after admission, neurobehavioral testing revealed normal orientation, attention, language, constructional abilities, calculation, reasoning, and judgment with impairment in verbal learning.

Surprisingly, the patient repeatedly expressed strong sensations of having previously known many of the hospital personnel. Sometimes he would ask if he knew them from school or his hometown, a rural village. The patient recognized that his feelings of familiarity were incorrect and strange but he continued to report the presence of these feelings. His sense of hyperfamiliarity for strangers gradually disappeared over a 2-week period, and he was discharged fully recovered.

Comment

The present case provides further evidence for the "assoziierende Erinnerungs- falschungen" phenomenon, or the sensation that unknown people are already known, originally described by Emil Kraepelin.[1] This person hyperfamiliarity syndrome is distinct from Fregoli's phenomenon, in which strangers are identified as familiar persons.[2] Fregoli patients change the personal identities of surrounding persons, but this patient only felt that they were familiar and did not change their identities.

This patient had a focal lesion in the left ACC, or Brodmann's Area 24. This corresponds to the "affect" region of the ACG, which regulates emotional awareness, as well as motivation and intention. Consistent with this localization, this patient had an initial akinetic muteness. His subsequent hyperfamiliarity syndrome implies that an alteration in emotional awareness for people is mediated by the ACC.

Prior investigations have suggested a right hemisphere person recognition network that links seen faces with representations of affective and personal relevance.[3–5] The right superior temporal gyrus, amygdala, and orbitofrontal cortex respond to facial emotions and contribute to a sense of familiarity from faces.[1–4] Hyperfamiliarity for unknown faces could arise from a hemispheric imbalance, with relative hypoactivation of left hemisphere processes but hyperactivation and spurious responsiveness of the right hemisphere person recognition network.[5]

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