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Late Onset Continuous Verbal Perseveration After Glial Tumor Surgery: A Rare Ictal Phenomenon

Glial Tümör Cerrahisi Sonrası Geç Başlangıçlı Verbal Perseverasyon: Nadir Bir İktal Fenomen

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ABSTRACT

OBJECTIVE: Perseveration is a rarely observed symptom during the course of neurosurgical diseases. As all perseverations are not uniform, determination of the corresponding brain regions is of great importance prior to deciding whether it represents a destructive or an irritative process.

CLINICAL PRESENTATION: A right-handed 33-year-old patient with a subtotally excised left temporoparietal low-grade astrocytoma was readmitted 9 months later with a complaint of new-onset continuous verbal perseveration which is primarily a sign of a non-dominant hemisphere abnormality. Repeat magnetic resonance imaging showed no change in the residual tumor size or in the edema. No new tumor focus was detected.

DISCUSSION: The patient's electroencephalogram indicated a contralateral right-sided epileptic focus. The carbamezapine dosage was increased and the perseverations subsided rapidly.

CONCLUSION: The late-onset ictal perseveration did not imply tumor progression and was triggered by the focus on the contralateral hemisphere, most probably caused by radiotherapy.

KEY WORDS: Glial tumor, ictal, radiotherapy, surgery, verbal perseveration

ÖZ

GİRİŞ: Nörolojik hastalıkların seyrinde motor perseverasyon nadir görülen bir bulgudur. Bütün perseverasyonların uniform olmadıkları göz önüne alındığında, destrüktif veya irritatif bir sürecin varlığı ve sorumlu beyin bölgesinin saptanması teşhis ve tedavi açısından oldukça önemlidir.

OLGU SUNUMU: Sağ el dominanslı 33 yaşında erkek hasta, sol temporoparietal yerleşimli düşük gradeli astrositomun subtotal eksizyonunu ve postoperatif radyoterapiyi takiben, postoperatif 6. ayda ani başlangıçlı, sürekli motor perseverasyon nedeni ile yatırıldı. Nondominant hemisfer kökenli bu klinik görünümün araştırılması sırasında uygulanan MR görüntülemede tümörün büyüklüğünde ve postoperatif 2. ayda saptanan radyonekroz alanının büyüklüğünde değişiklik saptanmadı. EEG tetkikinde sağ temporal epileptik odağı saptanan hastanın antiepileptik tedavisinin düzenlenmesini takiben perseverasyonun hızlıca kaybolduğu görüldü.

TARTIŞMA ve SONUÇ: Sunulan olguda, geç başlangıçlı motor perseverasyonun etyolojisi tümör progresyonu değildi. Nadir olarak görülen bu iktal fenomen düşünüldüğünde ve teşhis edildiğinde, antiepileptik tedavi ile başarılı sonuç alınabilmektedir.

ANAHTAR SÖZCÜKLER: Cerrahi, glial tümör, iktal, radyoterapi, verbal perseverasyon

INTRODUCTION

Perseveration is an inappropriate continuation or repetition of a response or activity and is thought to be pathognomonic of brain damage. Perseveration manifests itself in several different forms and many hypotheses have been proposed to explain the mechanisms underlying this abnormal behavior (3, 4). There are three forms of perseverative behavior each with its own anatomic correlation: recurrent perseveration is the repetition of a previous response to a subsequent stimulus, stuck-in-set perseveration is the inappropriate maintenance of a category of activity, and continuous perseveration is an abnormal prolongation of a current activity (5).

CASE REPORT

A 33-year-old right-handed male was admitted to the neurosurgery clinic with headaches localized to the left hemicranium for one month. He had experienced a generalized tonic-clonic seizure during sleep five days before admission. His neurological examination revealed no deficit. Cranial MRI demonstrated a solid tumor in the left temporal lobe (Figure 1). The tumor was subtotally excised via a left frontotemporal craniotomy and histopathological examination revealed a low-grade astrocytoma. Postoperatively, the patient underwent radiotherapy of 55 Gray in 30 fractions. Following the completion of radiotherapy, the patient was discharged 2 months after surgery without neurological deficit.

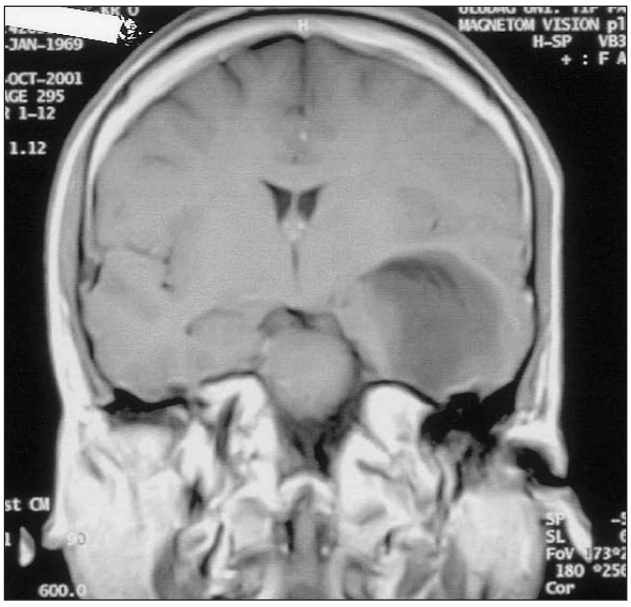

Figure 1: Coronal T1-weighted image shows the tumor in the left temporal lobe

Despite antiepileptic medication (diphenyl hydantoin 300 mg/day), a complex partial seizure occurred 4 months after the operation. The neurological examination after the seizure was found to be normal but phasic sharp wave activities and short paroxysms composed of slow waves with delta rhythm were recorded at the right temporal region. Previously prescribed phenytoin was replaced with carbamazepine (400 mg/day). After a 5-month seizure-free period, he was readmitted with complaints of agitation, aggressive behavior against himself as well as those around him and continuously repeating words during current speech. There was again no lateralized motor or sensory deficit, but there was disorientation to place and time, loss of initiation, apathy, agitation, motor and verbal perseveration, and acalculia. He exhibited non-fluent speech and his auditory comprehension was fair with some difficulty in comprehending complex commands. Naming was preserved and he was able to repeat words. Reading comprehension and writing were also intact. The most significant characteristic of his speech was continuous verbal perseveration. During free conversation and the examination, he often repeated phrases that consisted of his answers to the questions. However, he did not have any tendency to repeat the words spoken to others. He was aware of his repetitive behavior and was embarrassed by it.

Repeat cranial MRI demonstrated a residual mass in the left temporoparietal region and radiation necrosis but there was no increase in the size of the mass or edema (Figure 2). However, there was diffuse cortical atrophy and ventricular dilatation (Figure 3). EEG revealed phasic sharp wave activity and short paroxysms composed of slow waves with delta rhythm at the right temporal region.

His symptoms and signs were carefully evaluated and the preliminary diagnoses were: 1) right temporal epileptic activity with a rare clinical presentation 2) destruction or irritation of dopaminergic pathways in bilateral deep frontal and temporal lobes due to radiotherapy 3) microprogression of the tumor tissue effecting the dopaminergic pathways. A two-step treatment was planned. Taking the possibility of a rare form of epilepsy into consideration, the first step was to increase the dosage of antiepileptic therapy or change the antiepileptic drug. In case there was no response to antiepileptic drugs, the possibility of

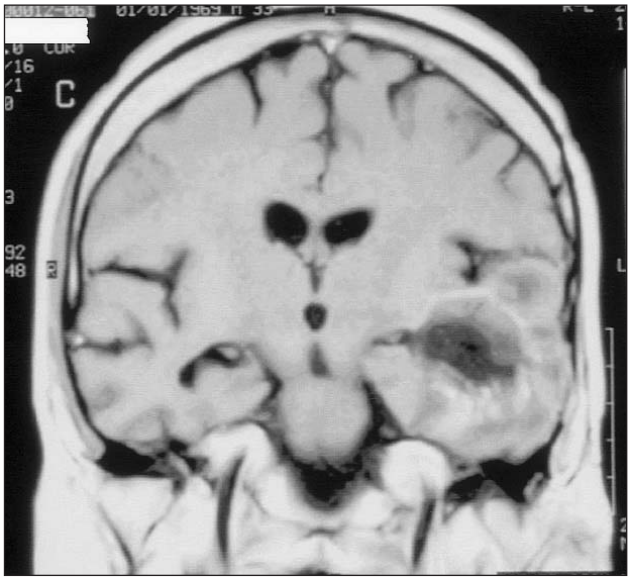

Figure 2: Post-contrast T1-weighted coronal image obtained 9 months after partial resection and radiotherapy. The intensity of the tumor has decreased. Note peripheral contrast enhancement that likely represents radiation injury rather than tumoral activity. The ventricles and the subarachnoid spaces have enlarged in the interval, reflecting diffuse cerebral atrophy probably due to radiation and/or chemotherapy injury

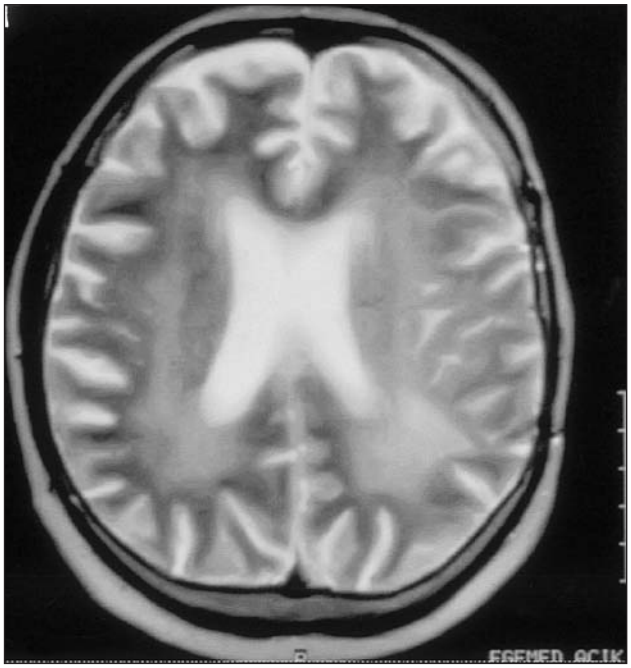

Figure 3: Axial T2-weighted image far above the tumor, obtained at the same time as Fig 2. Diffuse demyelination in both corona radiata indicates radiation injury. Enlargement of the ventricles and the subarachnoid spaces are also observed in this image.

destruction of the dopaminergic pathways would be the possible diagnosis and the second step of the therapy would focus on dopaminergic therapy solutions that have no known clinical applications.

As the dosage of carbamazepine was increased to 600 mg/day, the perseverative speech disappeared. The control EEG revealed no epileptic focus. The patient was discharged without any neuological deficit. The most recent examination was performed 2 years after discharge without any pathological findings.

DISCUSSION

Perseverative behavior is a feature of several neurological and psychiatric disorders, such as brain tumors, Alzheimer's disease, subcortical vascular dementia, anoxic brain injury, closed head injury, complex partial seizures, autism, Tourette syndrome, and schizophrenia (2, 6). Disruption of specific anatomic and pharmacologic (especially dopaminergic) systems produces different forms of perseveration that, in turn, underlie particular neurobehavioral disorders (8, 9, 10).

The approximate brain regions associated with perseverative behaviour have been identified, but some studies have demonstrated these sites in more detail. In a study of Sandson et al (9) three groups of patients (with aphasia, right hemisphere damage and Parkinson's disease) were evaluated to elicit the categories of perseveration. The authors proposed a theory of perseveration dependent on anatomic, neuropsychological, and pharmacologic factors related to cerebral dominance. The patients with aphasia produced significantly more recurrent perseveration than patients with right hemisphere damage or healthy controls. Stuck-in-set perseveration was associated with dopaminergic system dysfunction while continuous perseveration was related to right hemisphere damage (3, 4, 5, 10).

Although verbal perseveration is a rare symptom, the differential diagnosis with echolalia should be made. Echolalia is defined as the continuous repetition of the phrases that are directed to the patient. Echolalic patients also have a tendency to respond to the stimuli directed to other patients, so called "response-to-next-patient-stimulation". Echolalia is associated with medial frontal lesions in the language dominant hemisphere (4, 5).

Although the tumor was located in the left temporoparietal region, our patient's

symptomatology as well as the absence of the response-to-next-patient-stimulation sign indicated the presence of continuous perseveration rather than echolalia. Initial right-sided abnormal EEG discharges also supported this diagnosis.

Perseverative behaviour can be observed as a part of complex partial seizures as well as due to various parenchymal lesions (1). There are also some studies reporting visual and auditory perseveration as an epileptic phenomenon during complex partial seizures (7).

In our patient, the epileptic focus in the right temporal region aroused suspicion of the perseverative speech being an epileptic phenomenon. This focus might be explained by radiotherapy causing right temporal lobe gliosis. The EEG findings and perseveration subsided when the dosage of the antiepileptic medication was increased.

CONCLUSION

The late-onset ictal perseveration did not imply tumor progression and was triggered by an epileptic focus in the contralateral hemisphere most probably caused by radiotherapy.

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