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CASE REPORT / OLGU SUNUMU

Eating Epilepsy After Resection of Frontal Meningioma: A Case Report

Frontal Meningioma Rezeksiyonu Sonrası Yemek Yeme Epilepsisi; Olgu Sunumu

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Summary

Eating epilepsy (EE) is a rare type of reflex epilepsy. Eating-induced seizures are, in general, localization-related, and most commonly of the complex partial type. Less frequently, these seizures are of the simple partial type with or without secondary generalization. Here, we report a case of a 38-year-old female patient with EE after frontal meningioma resection. She had suffered from secondary generalized seizures unresponsive to any antiepileptic therapy for 3 years. She had both reflex and non-reflex seizures. However, most of the seizures were triggered by food or water deglutition. This was an unusual case of EE with secondary generalized tonic-clonic seizures.

Key words: Eating epilepsy; frontal meningioma resection; generalized tonic-clonic seizures.

Özet

Yemek yeme epilepsisi refleks epilepsinin nadir bir tipidir. Yemeyle tetiklenen nöbetler genellikle lokalizasyon ilişkilidir ve sıklıkla kompleks parsiyel tiptedir. Daha az sıklıkta, bu nöbetler sekonder jeneralize olan ya da olmayan basit parsiyel tiptedir. Bu yazıda, frontal menengioma ameliyatı sonrası gelişen, yemek yeme epilepsisi olan 38 yaşında kadın hasta sunuldu. Hasta, üç yıldır hiçbir antiepileptik tedaviye yanıt vermeyen sekonder jeneralize nöbetlerden yakınıyordu. Refleks ve refleks olmayan nöbetleri mevcuttu. Ancak çoğu nöbeti yemek yeme veya su içme ile tetikleniyordu. Bu olgu yemek yeme epilepsisinin sekonder jeneralize tonik klonik nöbetlerle giden nadir görülen bir formudur.

Anahtar sözcükler: Yemek yeme epilepsisi; frontal meningioma rezeksiyonu; jeneralize tonik klonik nöbetler.

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Introduction

Reflex epilepsy (RE) is characterized by seizures precipitated by an identifiable factor or external stimulus. REs are classified into two types as simple and complex. Simple RE is precipitated by simple sensory stimuli such as flashes of light or being startled, whereas complex RE is precipitated by complex or more elaborate stimuli such as specific pieces of music or eating.[1]

Eating epilepsy (EE) is a rare type of RE.[2,3] The majority of those patients present with both reflex and non-reflex seizures, although patients with reflex eating seizures alone may exist.[2,4] Eating-induced seizures are most commonly of the complex partial type. Less frequently, these seizures are of the simple partial type with or without secondary generalization.[5-7] Tonic-clonic motor manifestations[8] and periodic, brief tonic seizures of the limbs or neck are also reported as clinical features of EE.[6]

EE has also been described according to its trigger area, which is essentially of two types: suprasylvian or temporolimbic.[3] In patients with temporolimbic-onset seizures, taste and emotional and autonomic components of eating play an important role in the nature of the seizure trigger. However, suprasylvian seizures are induced by proprioceptive or somatosensory stimulation.[8] Hyperexcitability of the temporolimbic area involves susceptibility to gustatory, olfactory, affective, and emotional stimuli.[1,7]

Extralimbic (suprasylvian) regions have been implicated when the abnormal cortex is in a proprioceptive region and involves other sensory afferents (lingual, buccal or pharyngeal). These areas are activated by extensive sensory input generated by the complex behaviors involved in eating action.

Patients with seizures exclusively or mostly precipitated by eating make up 0.5 to 1 per 1,000 epileptic patients.[5] Most reports have identified the etiology to be associated with malformations of cortical developmental hypoxic brain injury, previous meningoencephalitis, or static encephalopathy.[1] Recently, other conditions, including familial or sporadic lateral temporal epilepsy, were also identified as playing roles in the EE etiology.[9]

We report a case of a 38-year-old female patient who developed EE after frontal meningioma resection. Most of the seizures were triggered by food or water deglutition. This was an unusual case of EE with secondary generalized tonic clonic seizures.

Case Report

A 38-year-old right-handed female patient presented to the emergency department because of frequent secondary generalized tonic clonic seizures. Diazepam (10 mg intravenously [IV]) was ineffective and the seizures, lasting about 3 to 4 min continued with hourly intervals. Subsequent sodium phenytoin (20 mg/kg in 500-mL isotonic solution) infusion stopped the seizures, and the patient had no postictal confusion. Cooperation was maintained, and her neurological examination was normal She was then transported to the neurology clinic. Computed tomography (CT) of the brain revealed changes indicating encephalomalacia in the right frontal area. Sodium valproat (1000 mg/day orally) was chosen for therapy. However, the seizures restarted a few days later. Clinical observation showed that almost all seizures were triggered by water and/or food deglutition; smelling or tasting food did not trigger the seizures. We observed three secondary generalized seizures shortly after drinking water or swallowing food, especially in the morning. When the oral dose was not of benefit, IV sodium valproat infusion (30 mg/kg/day) was administered, and the seizures ceased and did not recur during hospitalization. Blood valproic acid levels were in the normal ranges.

The patient reported that she also had less frequent non-reflex seizures (of approximately five seizures per month, one of them was a non-reflex seizure). Semiological events were similar for all seizures; as versive deviation of the head to the left side followed by generalized tonic clonic phenomena. Patient reported no subjective experience preceding the seizure. Clinical picture of our patient suggested EE. MRI revealed a 4-x2-x3-cm lesion featuring postoperative cystic encephalomalacia in the right frontal lobe and superior and middle frontal gyrus (Figure 1) secondary to the resection of frontal meningioma performed in 2008. Seizures were reported to begin six to seven months after the operation and were present for 3 years at the time of admission. Before then, the patient had no seizures and the sole manifestation of the brain lesion was headache.

Despite frequent seizures, EEG of the patient during resting revealed no abnormality; however, some sharply contoured slow waves were prominent on right frontal regions which

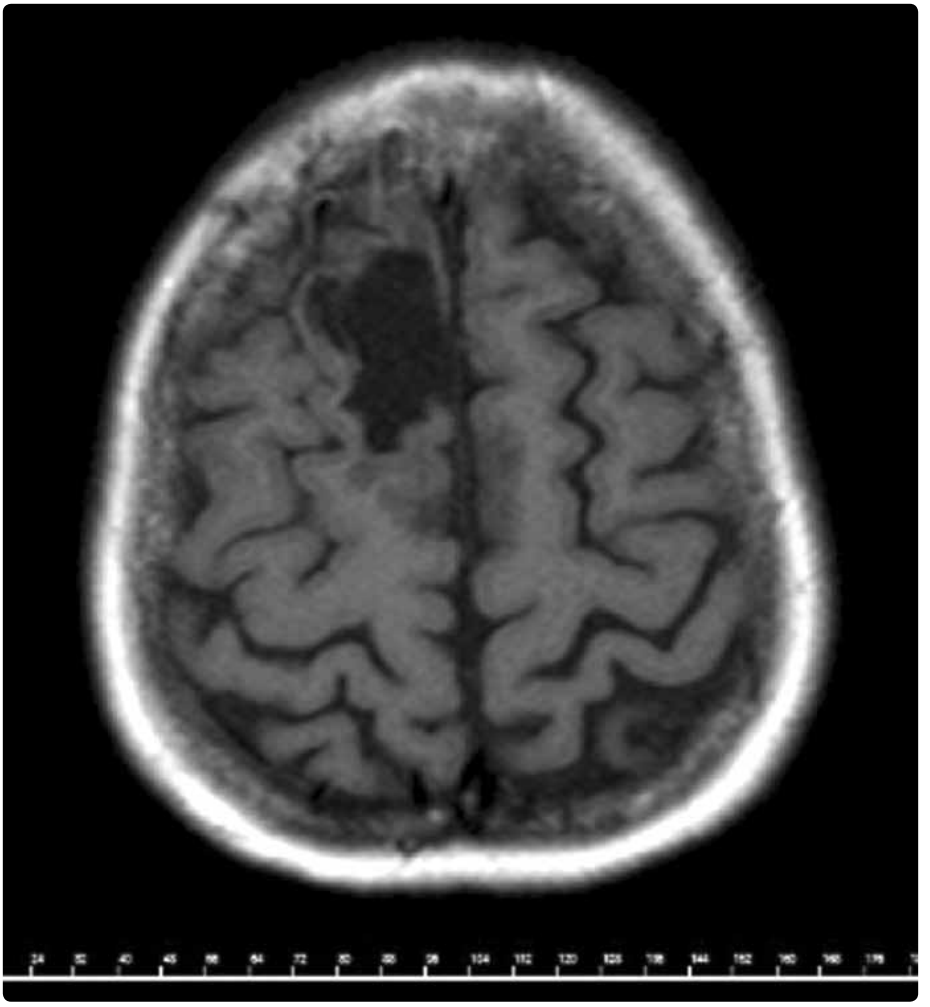

Figure 1. MRI revealed a 4 × 2 × 3 cm postoperative cystic encephalomalacic area in the right frontal lobe and superior and middle frontal gyrus.

spread to other regions, during hyperventilation. Depending on the electro-clinical and imaging data of the present case, the diagnosis of reflex EE originating from the right frontal lobe with widespread diffusion was reinforced.

There was no family history of either epilepsy or febrile convulsions. Although many different antiepileptic agents (topiramate, levetiracetam, phenytoin sodium, and sodium valproate) were tried, her seizures could not be controlled. She could not take the therapeutic doses of these drugs orally, because of reflex seizures.

Oral sodium valproat at 500 mg twice per day was prescribed after patient's discharge from the hospital. Throughout a 6-month follow-up period, she had one generalized seizure following food deglutition, and levetiracetam at 500 mg twice per day was added to the therapy.

Discussion

The mechanisms of reflex EE are poorly understood. Wieser's critical mass theory proposes that complex RE occurs in response to a stimulus that triggers a "critical mass" of cortex by recruiting increased amounts of epileptogenic neurons. [10] On the other hand, the sensations of smell and sight, the taste of food, gastric distension, hyperglycemia, the masticatory movements of the jaw, tongue action, and swallowing have all been suggested as triggering factors. However,

those factors are not sufficient for satisfactory explanations for the pathophysiology of EE which remains unclear yet.[2,7] Nevertheless, the majority of authors postulate that multiple precipitating factors are involved in the process, or the complete sequence of eating a meal is required to provoke a seizure.[4,5]

In our case, the seizures started following chewing, swallowing or drinking. Masticatory movements of the jaw, tongue action, or swallowing may have been the triggering factors. The smell or taste of food did not trigger seizures. Available clinical and laboratory data strongly suggest that the epileptojenic zone is related to the post-operative lesion in the right frontal area. This suprasylvian area was reported to be related to EE with secondary generalization.[8] Despite the evidences suggesting temporal or extratemporal involvement in the production of eating-induced seizures, interictal patterns reveal that the seizures may not originate solely from one area and can be bilateral, unilateral, or even originate in the brain stem.[8] Thus, the epileptogenic focus and the seizure type are not always related to each other. Another theoretical assumption for the pathophysiology of this type of epilepsy is that, it may originate from any region of the brain. The amygdala has been suggested as a likely target for chronic and repetitive oral and masticatory actions implicated in EE.[8] The temporolimbic and insular areas were suspected to be crucial in generating eating seizures at the cortical level, but those episodes are triggered by peripheral input from organs innervated by the vagus nerve. It is possible that the unilateral vagus nerve bilaterally modulates the ascending activity at the brain stem level, after going through the tractussolitarius complex.[4] In addition to all these mechanisms, Blauwblomme et al.[11] identified overlaps between the gustatory cortex, involving insular, parietal, and frontal regions, and a frontoinsulo-hippocampal epileptogenic network. This epileptogenic network theory may support the mechanism of EE after frontal meningioma resection in our patient. However, the pathways involved between the peripheral receptor sites and the specific epileptogenic area in this case, are not available for speculatory considerations since the cerebral lesion is extensive and no specific epileptogenic site is recognized, no ictal EEG could be performed, as also the seizure triggering activities were variable as chewing, drinking water and swallowing food.

To our knowledge, this is the first report of EE with secondary generalized tonic clonic seizures after frontal meningioma surgery. A patient similar to ours was described by Auvinet al.[12] in 2010. They described a 7-year-old patient with reflex late-onset spasms (LOS) induced by water deglutition. That special case of reflex LOS was secondary to gliosis after surgery for a meningioma, as was the case in our patient. This case also involved suprasylvian EE due to frontal meningioma resection without lateralizing signs and no secondary generalization. As demonstrated by the present case, EE seems to involve complex interactions between frontal lobe centers and peripheral receptor organs in the oro-pharyngeal cavity and may present with apparently generalized motor seizures resistant to conventional therapy.

Acknowledgement

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines. This text English was corrected professionally by native speaker English consultants.

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