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Unilateral opercular lesion and eating-induced seizures

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ABSTRACT – Eating-induced seizures are an uncommon presentation of reflex epilepsy, a condition characterized by seizures provoked by specific stimuli. Most reports have identified aetiology associated with malformations of cortical developmental, hypoxic brain injury, previous meningoencephalitis or static encephalopathy. We present a patient with eating-induced reflex seizures, which began several years after treatment for an opercular primitive neuroectodermal tumour (PNET), and who subsequently underwent in-depth clinical and video-EEG analysis for her seizures. This patient noted rapid improvement with decreased frequency of seizure activity after treatment with valproic acid. We discuss the aetiology of reflex epilepsy, the anatomical basis of eating-induced epilepsy, and review the current literature. *[Published with video sequences]*

Key words: reflex epilepsy, eating seizures, eating epilepsy, video-EEG monitoring, operculum

Reflex epilepsy is characterized by seizures precipitated by an identifiable factor or external stimulus. They are classified as two types; simple or complex. Simple reflex epilepsy is precipitated by simple sensory stimuli such as flashes of light or by being startled, whereas complex reflex epilepsy is precipitated by complex or more elaborate stimuli such as specific pieces of music or eating. Although the seizures seen in patients with reflex epilepsy may occur with partial or generalised onset, seizures in relation to food are almost exclusively related to symptomatic focal epilepsy (Zifkin and Andermann, 2006). Most reports have identified aetiology associated with malformations of cortical developmental (Verdu and Ruiz-Falco, 1991; D'Orsi *et al.*, 2007; Kishi *et al.*, 1999), hypoxic brain injury (Loreto *et al.*, 2000), previous meningoencephalitis (Mateos *et al.*, 1995), or static encephalopathy (Labate *et al.*, 2005). We describe the imaging and video-EEG data of a patient with a history of treated opercular primitive neuroectodermal tumour (PNET) who developed eating-induced seizures.

Case report

A 23-year-old right-handed Caucasian woman with a history of previous PNET was admitted to the hospital for evaluation of paroxysmal stereotyped episodes of head dropping

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upon eating. At 30 months of age, she began to preferentially use her left hand, and was noticed to have weakness of the right arm and subsequent ataxia. Diagnostic evaluation revealed a left opercular PNET. She first underwent tumour resection and received chemotherapy, followed nine months later by whole brain and spine irradiation. Subsequent periodic brain imaging did not reveal recurrence, but she was left with weakness of the right arm and a mild static encephalopathy. At approximately eight years of age, she began having stereotyped attacks described as loss of head control. Her head would fall forwards and occasionally be accompanied by a jerk of the right arm. The episodes were not associated with other neurological symptoms. The attacks occurred approximately once a year, but increased at the age of 10 to a frequency of once a month. Numerous antiepileptic drugs (AEDs), including carbamazepine, lamotrigine, topiramate, and levetiracetam did not control the events.

The association of seizures with eating developed during adolescence. At the age of 23, the seizures began to occur exclusively with every meal, with multiple events being provoked during a single meal and each seizure lasting less than five seconds in duration.

She was admitted to the epilepsy monitoring unit at St Luke's Episcopal Hospital in Houston, Texas in order to characterize these events. Attempts to induce episodes were made by placing food in front of her so she could smell the aroma, asking her to make chewing movements without food in her mouth, to drink liquids, to think about food while it was not there, and keep food in her mouth without chewing. None of these manoeuvres induced the events, but multiple episodes occurred upon the act of eating during each meal. The meals consisted of hospital food such as eggs, pancakes and sausages in the morning. For lunch and dinner meals predominantly consisted of meat or poultry. Occurrence of the seizures was independent of the timing of the meals and the type of food.

Inpatient video-EEG monitoring recorded stereotyped episodes of a spontaneous head drop with eyes open, accompanied by brief myoclonus of the right arm, intermittently accompanied by oral myoclonus *(see video sequence)*. Concurrent electromyography (EMG) was not performed as part of the routine video-EEG monitoring, thus excluding definitive confirmation of atonia. The patient answered questions appropriately and followed commands during the cluster of seizures. Clusters would include 15-20 per meal and lasted approximately 15 minutes. Inter-ictal EEG revealed continuous left hemispheric slowing, with predominantly moderate to high voltage (60 to 110 μV) polymorphic theta and delta activity. Frequent high voltage (100 to 120 μV) sharp waves were seen over the left temporal area, with shifting points of an area of maximum electronegativity over the scalp between the left mid-temporal and posterior temporal areas, maximal over T7 and P7, respectively *(figure 1)*. Ictal EEG consisted of high voltage (90 to 110 μV), broadly-distributed, frontally-predominant delta activity, intermittently admixed with low-voltage fast activity *(figure 3)*.

The patient was examined by MRI at the age of 23, one week prior to admission to the epilepsy monitoring unit, which depicted post-treatment changes, as shown in *figure 2*. There was no change in MRI, based on MRI she had previously undergone since the age of eight.

The patient's AED regimen was changed from a combination of topiramate and lamotrigine to valproate and lamotrigine polytherapy. After a two-month follow-up period, she described a marked reduction in her seizure frequency with seizure-freedom during most meals.

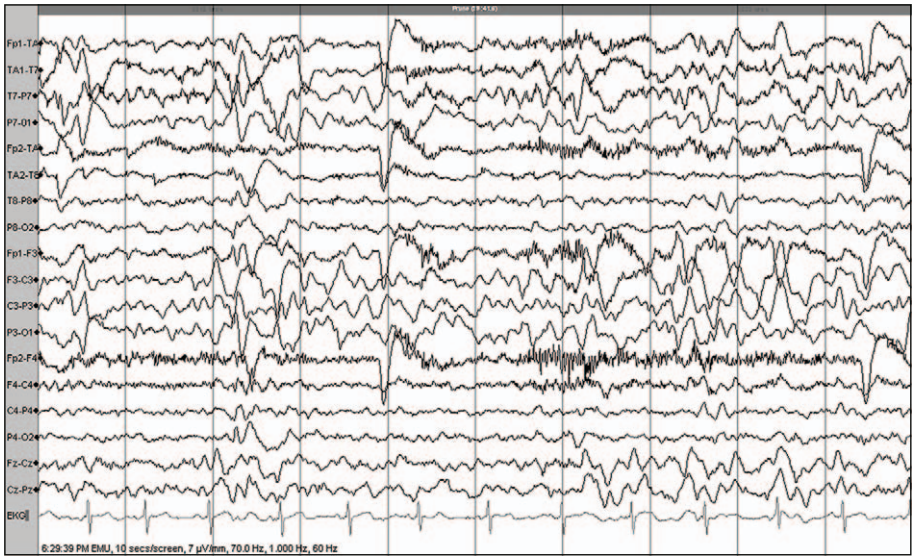

Figure 1. Interictal EEG.

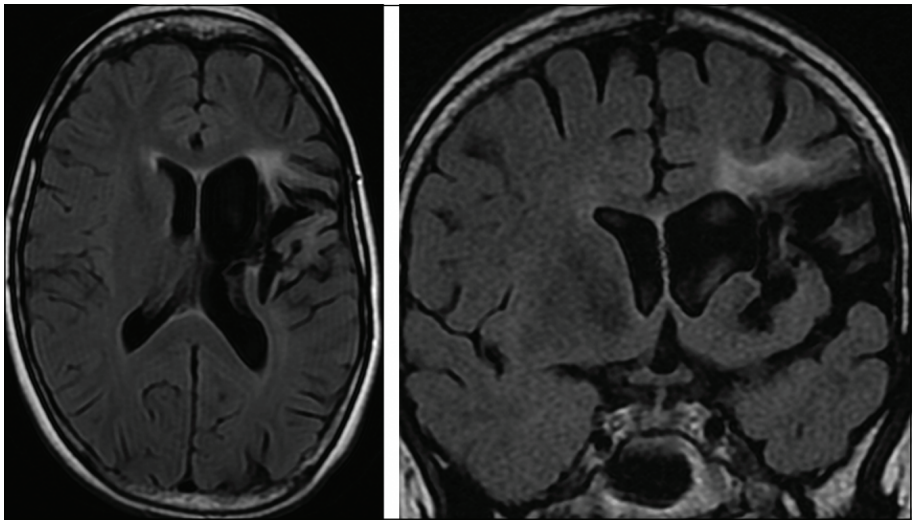

Figure 2. Axial and coronal FLAIR MRI.

Discussion

The mechanisms of reflex eating epilepsy are poorly understood. Multiple theories have been proposed over the years. Wieser's critical mass theory proposes that complex reflex epilepsies occur in response to a stimulus that triggers a "critical mass" of cortex by recruiting increased amounts of epileptogenic neurons (Wieser, 1998). Functional imaging studies have shown that reading epilepsy involves speech and language networks in both hemispheres. Several studies have reported that the critical mass is functionally linked (Rémillard *et al.*, 1998). In eating epilepsy, proposed triggering mechanisms include mastication (Scollo-Lavizzari and Hess, 1967), oesophageal stimulation (Forster, 1971), and the satisfactory feeling associated with eating (Cirignotta *et al.*, 1977). There is significant aetiological heterogeneity associated with several pathophysiological mechanisms which underlie eating epilepsy (Rémillard *et al.*, 1998; Fiol *et al.*, 1986). Some investigators have suggested the interaction between temporolimbic and extratemporal regions is responsible for eating epilepsy (Fiol *et al.*, 1986). Hyperexcitability of the temporolimbic area involves susceptibility to gustatory, olfactory, affective, and emotional stimuli. Eating is suggested to induce constant activation in patients with temporolimbic seizures (Rémillard *et al.*, 1998) and these patients show reflex eating epilepsy from onset and continue to have most of their seizures with meals. Extralimbic (suprasylvian) regions have been implicated when the abnormal cortex is in a proprioceptive region and involves other sensory afferents (lingual, buccal, pharyngeal). These areas are activated by extensive sensory input generated by the complex behaviours involved in eating (Rémillard *et al.*, 1998). Indeed, our patient and others cited in the literature (Nakazawa *et al.*, 2002; Labate *et al.*, 2005; D'Orsi *et al.*, 2007) had lesions involving the frontal operculum. A summary of recently well-studied cases of eating-induced seizures is presented in *table 1*. As the table demonstrates, despite the suggestion of temporal or extratemporal involvement in the production of eating-induced seizures, inter-ictal patterns reveal that the origins of the seizures may not originate solely from one area and can be bilateral, unilateral or even originate in the brainstem (Nakazawa *et al.*, 2002). Thus, the mechanisms involved in producing these seizures are complex and diverse. While mechanisms may be complex, the clinical seizure types appear to be related to the localization of the relevant lesion(s), whatever their aetiology, as is usual for focal onset reflex seizures of all types.

Interestingly, our patient's electroclinical picture is consistent with that seen in patients with both infantile and late onset spasms. The high voltage transient diffuse slow-wave activity followed by electrodecremental activity, as seen on the ictal EEG in our patient, is similar to the pattern present in over a third of patients with infantile spasms (Kellaway *et al.*, 1979; Fusco and Vigevano, 1993). These EEG findings have also been reported to occur in tonic seizures (Chatrian *et al.*, 1982).

Previous studies have described patients with focal lesions that led to epileptic spasms (Labate *et al.*, 2005; Nakazawa *et al.*, 2002). Auvin *et al.* (2010) suggested that in order for an epileptic spasm to occur in older children, a wide epileptogenic area should be present. Late-onset spasms (LOS) have been described previously with infantile epileptic encephalopathy (Auvin *et al.*, 2010, Nordli *et al.*, 2007). Patients similar to ours have been described with reflex-induced epileptic spasms in response to water deglutition, following the resection of a meningioma (Auvin *et al.*, 2010).

Important limitations of our study should, however, be noted. Firstly, the exact mechanism underlying the head

Table 1. Summary of clinical, imaging, and electroencephalographic features of recent case reports.

Age	Gender	Etiology	Semiology	Association with Food	Interictal EEG	Ictal EEG	MRI	Outcome	Source
32	M	TLE	Déjà vu, complex partial seizures	During meals	Right temporal sharp waves	Right antero-mesial onset	Not reported	Seizure free following right temporal resection	Remillard, *et al.*
46	F		Hemi-sensorimotor	During meals		Right frontocentroparietal onset	Not reported	90% seizure reduction following right fronto-parietal resection	Remillard *et al.*
20	M	Viral meningoencephalitis	Focal motor seizure or the left face	During meals	Right centroparietal interictal discharges	Not recorded	Opercular atrophy	Reduction of seizures with polytherapy	Mateos *et al.*
25	M	Polymicrogyria	Atonic	During meals	Bilateral synchronous centroparietal sharp waves	Diffuse attenuation	Bilateral perisylvian polymicrogyria	Poor response on polytherapy	Kishi *et al.*
32	M	HIE	Atonic	During meals, sight of food, occasionally spontaneous	Bilateral independent sharp waves	Diffuse slowing then attenuation	Left ventriculomegaly	Reduction of seizures with VPA and LTG	Loreto *et al.*
23	F	HIE	LOC, left head deviation, oral automatisms	Five minutes after meals		Initial diffuse electrodecrement	Hyperintesnity in right retrotrigonal area	Poor response on polytherapy	Loreto *et al.*
42	M	HIE	LOC, focal motor, and automatisms	Start of lunch, sight of food	Right temporal sharp waves	Not recorded	Normal	Reduction of seizures on polytherapy	Loreto *et al.*
8	M	LGS	Atonic	Two minutes after eating a full meal	Diffuse slowing, slow spike and wave	High voltage diffuse slow spike and wave complex	Normal	Reduction of seizures with VPA	Lee *et al.*
11	M	Cryptogenic	Tonic spasms	During meals, swallowing without food	Right frontal sharp waves	Diffuse high voltage delta	Normal	Poor response on polytherapy	Nakazawa *et al.*
8	M	Microcephaly, global developmental delay	Tonic spasms	During meals	Generalized spike and polyspike wave	Diffuse spike and wave followed by diffuse attenuation	Poorly formed and thickened opercular region	Reduction of seizures with VPA	Labate *et al.*
26	M	Microcephaly, global developmental delay	Tonic spasms	During meals	Right temporal slowing and sharp waves	Diffuse spike and wave followed by diffuse attenuation	Normal	Reduction of seizures with VPA and VGB	Labate *et al.*
30	M	HIE	Atonic	During meals	Left temporo-parietal sharp waves	Diffuse slow wave complex	Bilateral opercular dysplasia	Poor response on polytherapy	d'Orsi *et al.*
67	M	Medullary hemangioblastoma	Aura of metallic taste, ictal coughing and vomiting	During meals	Normal	Not described	Medullary hemangioblastoma	Not described	Rosenzweig *et al.*
34	M	Polymicrogyria	Temporal complex partial	During meals	Independent bitemporal spikes	Bilateral independent temporal onsets	Bilateral perisylvian polymicrogyria	Reduction of seizures with VNS	Cukiert *et al.*
40	F	Cryptogenic	Temporal complex partial	During meals, hot-water seizures	Independent bitemporal spikes	Temporal onsets	Normal	Reduction of seizures with VNS	Cukiert *et al.*
37	M	Cryptogenic	Temporal complex partial	During meals	Left fronto-central spikes	Left fronto-central onsets	Normal	Reduction of seizures with VNS	Cukiert *et al.*

M: male; F: female; TLE: temporal lobe epilepsy; HIE: hypoxic ischemic encephalopathy; LGS: Lennox Gastaut Syndrome; LOC: loss of consciousness; VPA: valproic acid; VGB: vigabatrin; LTG: lamotrigine; VNS: vagus nerve stimulator.

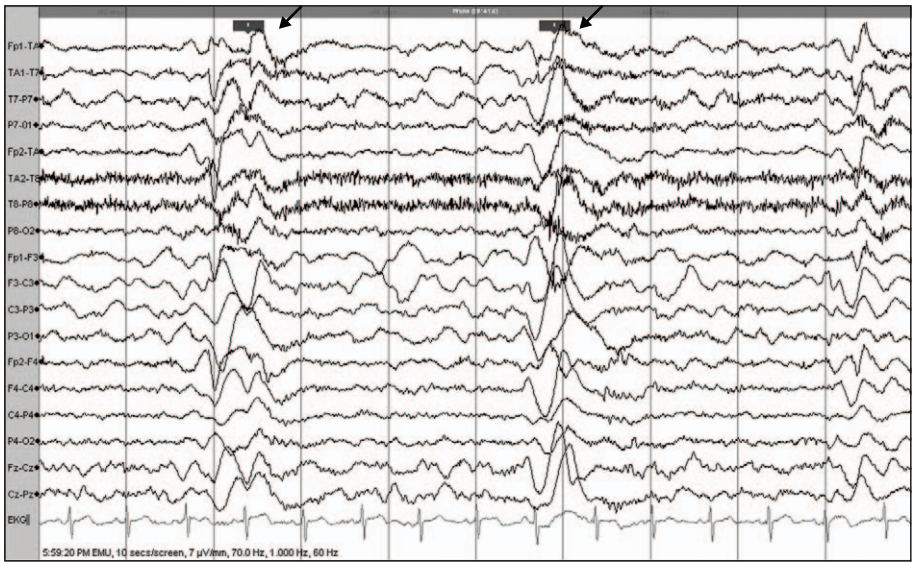

Figure 3. Ictal EEG. Seizures associated with diffuse delta (arrows).

drops could not be demonstrated since there was no ictal surface EMG polygraphic recording. Secondly, we are unable to discuss long-term prognosis since our patient had only recently undergone video-EEG monitoring. Nevertheless, this case study contributes to the growing literature and a clearer description of eating-induced reflex epilepsy. □

Legend for video sequence

Eating induced seizures.

Disclosure
None of the authors has any conflict of interest or financial support to disclose.

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