Clinical commentary

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New-onset musicogenic epilepsy after temporal lobe epilepsy surgery

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ABSTRACT – Musicogenic epilepsy is a reflex epilepsy provoked by listening to or playing music. The epileptogenic network involves temporal regions, usually mesiotemporal structures. We present a 31-year-old female patient who experienced musicogenic seizures after a right temporal lobectomy with amygdalohippocampectomy that was performed in order to treat preexisting right mesio-temporal epilepsy.

Key words: reflex epilepsy, temporal lobe resection, musicogenic seizures, epilepsy surgery

Musicogenic epilepsy is a rare but well-defined form of reflex epilepsy consisting of seizures that are consistently triggered by a specific auditory stimulus, varying from simple tones to complex melodies (Stern, 2015).

Temporal lobe involvement (both right and left lobes, respectively) in the epileptogenic network of musicogenic epilepsy is well documented (Wieser *et al*., 1997). Recent reports of the successful treatment of musicogenic seizures with anteromesial temporal lobe resections support the understanding that musicogenic epilepsy could be a limbic epilepsy (Stern, 2015).

Here, we present the occurrence of musicogenic seizures in a patient after a standard right temporal lobectomy with amygdalohippocampectomy was performed in order to treat a previously existing pharmacoresistant right mesiotemporal epilepsy.

Case study

A 31-year-old, right-handed woman had seizure onset at the age of 21. Since then, she experienced mainly diurnal seizures, consisting of a sense of fear, then unpleasant feeling in the stomach or chest which lasted several seconds, followed by a sense of heart beating. She usually stopped with current activities but without losing consciousness. Her mother who sometimes witnessed the seizures, described a sudden onset of motionless staring, followed by non-purposeful movements of the right hand or nose rubbing. She had two or three

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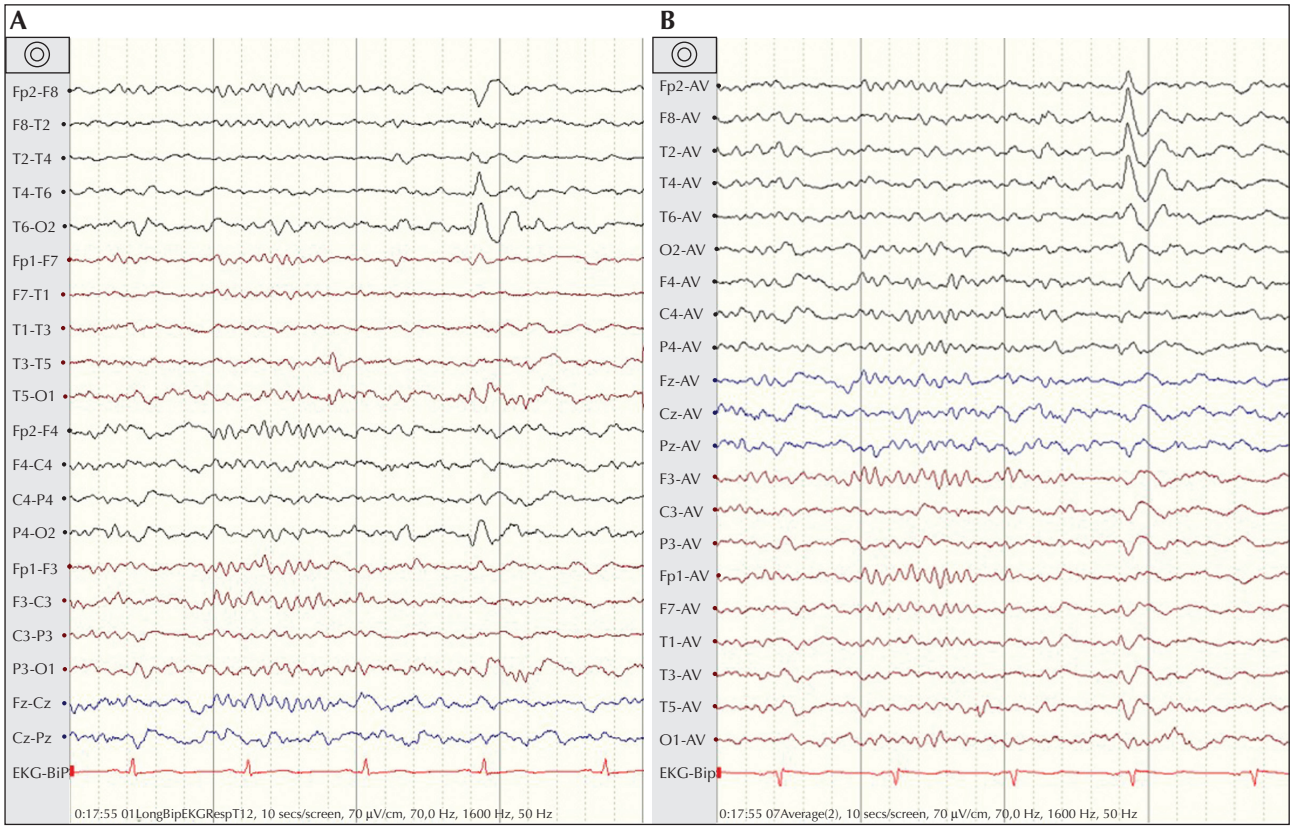

Figure 1. Preoperative interictal EEG discharges over the right temporal region during sleep in longitudinal bipolar (A) and referential (B) montages.

episodes per day and the seizure frequency remained unchanged despite two different antiepileptic drug regimens (levetiracetam and lamotrigine).

The history of the patient was unremarkable and she had no obvious epilepsy risk factors. The patient was not especially interested in music and had no specific musical education. Her family history was negative for epilepsy and other neurological diseases. Her neurological and physical status were unrevealing.

The patient's interictal EEG demonstrated frequent spikes and sharp waves over the right temporal region (*figure 1*). During long-term (96-hour) video-EEG monitoring, 10 habitual automotor seizures were recorded (sense of fear->right hand automatism->brief oroalimentary automatisms->left hand immobility with dystonic posturing-> right hand postictal nose wiping), all with ictal onset zone in the right temporal region (*figure 2*). Immediately after the seizure, she was able to speak.

Brain MRI revealed sclerosis of the right hippocampus, while interictal ^{18}F-FDG PET highlighted a hypometabolic zone of the right mesiotemporal region (*figure 3*). The neuropsychological assessment indicated mild disturbances in non-verbal memory.

A standard right temporal lobectomy with amygdalohippocampectomy was performed when the patient was 29 years old. The immediate postoperative course was uneventful aside from transient double vision limited to down gaze. Interictal EEG showed sporadic abnormal slowing across the right temporal region without any spikes or evolvement during the postoperative follow-up. A histopathological examination confirmed the existence of hippocampal sclerosis (ILEA type III; end-folium sclerosis).

The patient was seizure-free for three months after the surgical intervention, but then started to experience a new form of paroxysmal events despite full compliance with the antiepileptic drug regimen. The patient herself noted a particular type of music as the exclusive provocative factor; not all forms of music were equally likely to provoke a seizure. The music of the popular pop-folk performer, Aleksandra Prijović, was the trigger most likely to produce a seizure. No spontaneous postoperative seizures occurred.

The patient was admitted to the video-EEG telemetry unit for reassessment. Seizure-eliciting music, the song "*Za nas kasno je*" by Aleksandra Prijović, was played on an MP3 player until a cluster of two

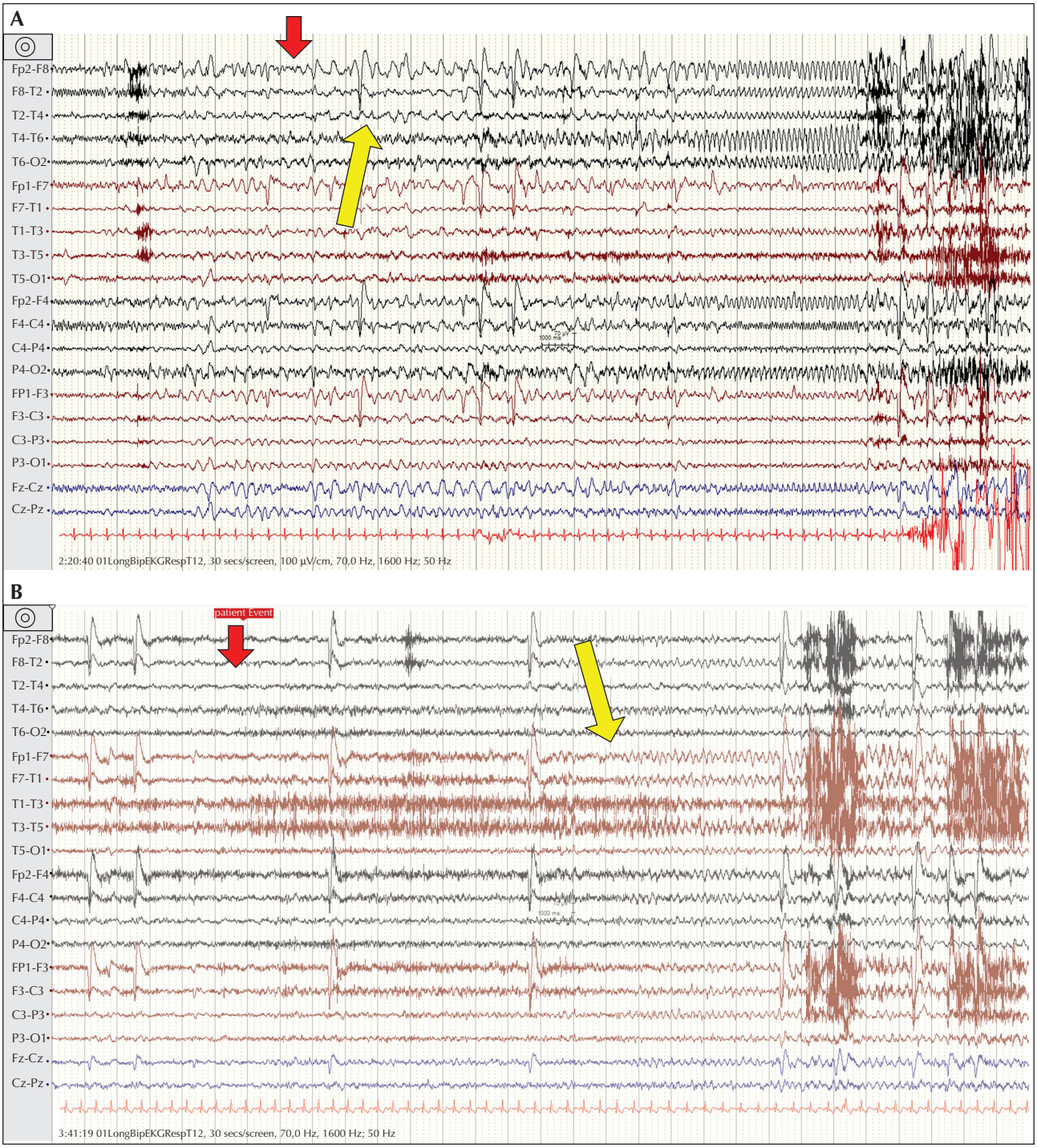

Figure 2. Preoperative ictal EEG onset in the right temporal lobe (A) and postoperative ictal EEG onset mainly over the left temporal lobe although an early but much less prominent involvement of the right temporal lobe can be noticed 180 seconds after the beginning of seizure-eliciting music (B).
Red arrows indicate occurrence of aura and yellow arrows point to ictal EEG activity. EEG format: 30 seconds/per page.

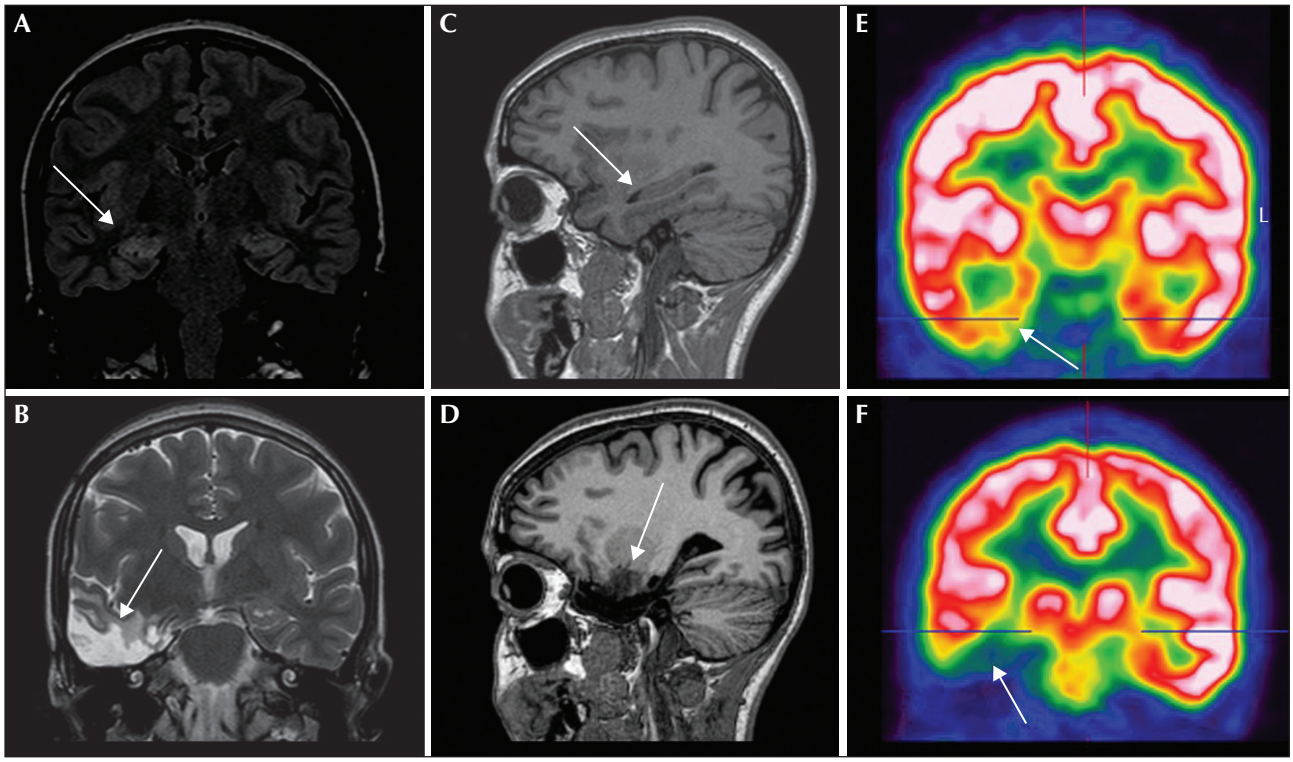

Figure 3. Coronal and sagittal planes of brain MRI and interictal ^{18}F-FDG PET showing preoperative right hippocampal sclerosis (A, C); preoperative hypometabolic zone in the right mesiotemporal region (E); postoperative sequel after standard right temporal lobectomy with amygdalohippocampectomy (B, D); and postoperative ametabolic zone in the right mesiotemporal region only (F) (the arrows in each panel refer to each respective finding).

electrographic seizures was recorded. Seizure semiology involved a sense of anxiety, then tachyarrhythmia (HR >120/min), and global aphasia. The ictal EEG activity was localized to the left temporal region, lasting up to 30 seconds. Postoperative brain MRI revealed only normal findings associated with a standard right temporal lobectomy with amygdalohippocampectomy. New interictal ^{18}F-FDG PET showed ametabolism at the site of the previous resection (*figure 3*). A neuropsychological reassessment indicated an existing disturbance in verbal memory and a declining trend in the field of visual memory.

Discussion

Musicogenic epilepsy is a well-known entity that continues to draw the attention of researchers as a potential model for examining the principles of epileptogenesis, but the dual and paradoxical relationship between music and epilepsy is intriguing and still unresolved (Tseng, 2018). Cases of preoperative musicogenic seizures replaced by other focal seizures have been reported (Hennessy, 2000). We present, to the best of our knowledge, a first case of postoperative musicogenic seizures.

The clinical features of our patient did not differ significantly from the clinical features of the cases reported so far: female gender, age in the twenties at epilepsy onset, spontaneous preceding musicogenic seizures, frequent autonomic and automotor manifestations, and temporal region involvement (Maguire, 2017). The trigger for our patient was a specific type of music. This is, by far, the most frequently described situation in the literature (Kaplan, 2003). The three-minute latency before seizure onset could be highlighting the cognitive and emotional aspects of the musicogenic trigger (Stern, 2015).

Altered seizure semiology after a resection could be a response to network reorganization or new-onset epileptogenesis. An ictal SPECT study confirmed the dominant role of the temporal lobes in musicogenic epilepsy and provided some evidence of the suspected participation of mesial temporal lobe structures (Wieser *et al.*, 1997). Patients with surgically refractory temporal lobe epilepsies are prone to structural network rearrangements that exhibit higher connectivity between regions within the ipsilateral medial and lateral temporal lobe regions, but also with the contralateral temporal pole (Bonilha *et al.*, 2013). Therefore, postoperative structural network

rearrangements could lead to new-onset postoperative musicogenic epilepsy.

The postoperative ictal EEG, revealing a left-side seizure onset, could be explained by the "burned-out hippocampus" phenomenon (right hippocampus severely damaged by the resection) due to the faster propagation of ictal rhythms to the opposite temporal lobe than to the ipsilateral one through the dorsal hippocampal commissure (Nair *et al*., 2017).

The fact that our patient had early seizure recurrence within three months after the surgery suggests an inadequate resection of the epileptogenic focus rather than a maturing epileptogenicity in a surgical scar (Najm *et al*., 2013). The later course of intractable epilepsy in our patient also favours this particular hypothesis.

Extratemporal lobe epilepsy mimicking temporal lobe epilepsy or temporal plus epilepsy could be alternative explanations for surgical failure, although they are less likely in this specific case (Harroud *et al*., 2012).

The question remains whether postoperative musicogenic seizures are a consequence of epileptic network rearrangement or a possible sign of new-onset epilepsy caused by surgical cortical injury. Given our current knowledge, the first alternative seems more reasonable. □

Disclosures.
None of the authors have any conflict of interest to declare.

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