A Rare Finding in Epilepsy: Ictal Singing and Spitting

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Abstract

Singing is an emotional musical vocalization, with several forms and styles in different cultures. Ictal singing in focal epilepsy is rare. It is accepted as a complex automatism, with only few reported cases. We reported a case with complex automatisms that included ictal singing and spitting, with an epileptogenic zone in the right temporal lobe. A 24-year-old right-handed man was referred for pharmacoresistant epilepsy. Despite administering three anticonvulsant drugs in efficient doses, seizures occurred several times per month. The seizures were characterized by staring for a few seconds, during which the patient stayed unresponsive. This was followed by spitting and singing, the same song each time. The song was from a television-drama series that he had watched during his childhood and had an emotional impact on him. Our patient sang the same song during every attack. The song was from a television series. The patient had watched the series during his childhood and was emotionally influenced with it. Therefore, it might be the release of subconscious emotions suppressed during childhood, which stimulated him to recall the song and sing it during the seizures.

Keywords: Automatism, epilepsy, singing

Epilepside Nadir Bir Bulgu: İktal Şarkı Söyleme ve Tükürme

Öz

Şarkı söylemek, farklı kültürlerde çeşitli biçimleri ve stilleri olan duygusal, müzikal seslendirme olarak tanımlanabilir. Fokal epilepsilerde iktal dönemde şarkı söylemek oldukça nadir gözlenir. Literatürde az sayıda vaka bildirilmiştir. Bu eylem, kompleks bir otomatizma olarak kabul görmektedir. Bu yazımızda, sağ temporal lobda epileptojenik bölge tespit edilmiş olan iktal şarkı söyleme ve tükürme davranışı gösteren bir olgu bildirmeyi amaçladık. Olgumuz 24 yaşında ilaca dirençli epilepsi tanısı ile epilepsi merkezimize yönlendirilmiş olan bir hasta; etkin süre ve dozlarda üç antikonvülsan ilaç tedavisi altında ayda birkaç kere tekrar eden nöbetler ile başvurdu. Nöbetler, hastanın davranışsal ve motor donma göstermesi ile başlıyor; ardından tükürme ve şarkı söyleme gelişiyordu. Hastamız her atakta aynı şarkıyı söylüyordu. Hastanın her nöbetinde söylediği bu şarkı, çocukluğunda izlediği ve üzerinde duygusal bir etkisi olan bir televizyon dizisinden geliyordu. Bu durumu, çocukluğu sırasında kendisini çok etkilemiş olan bu şarkının nöbetler sırasında serbestlenmesi olarak yorumladık.

Anahtar Kelimeler: Otomatizma, epilepsi, şarkı söylemek

Singing is an emotional musical vocalization, with several forms and styles in different cultures. Ictal singing in focal epilepsy is rare. It is accepted as a complex automatism, with only few reported cases [1-5]. Several studies have supported their justification by using the neuronal networks of normal singing [6, 7]; however, the mechanism underlying ictal singing remains unclear.

We reported a case with complex automatisms that included ictal singing and spitting, with an epileptogenic zone in the right temporal lobe.

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Case Presentation

A 24-year-old right-handed man was referred for pharmacoresistant epilepsy. Despite administering three anticonvulsant drugs in efficient doses (levetiracetam, carbamazepine, and valproic acid), seizures occurred several times per month. The patient started experiencing seizures at the age of 20 years, which were characterized by a sequence of staring, singing of the same song, and spitting, with no postictal confusion. His neurological examination revealed no abnormality. Moreover, neither the patient nor his family members revealed any remarkable story, and there was no family history of seizures. He did not have any history of musical experience or educational background.

Scalp video-electroencephalography (EEG) monitoring revealed interictal sharp waves in the right tempo-

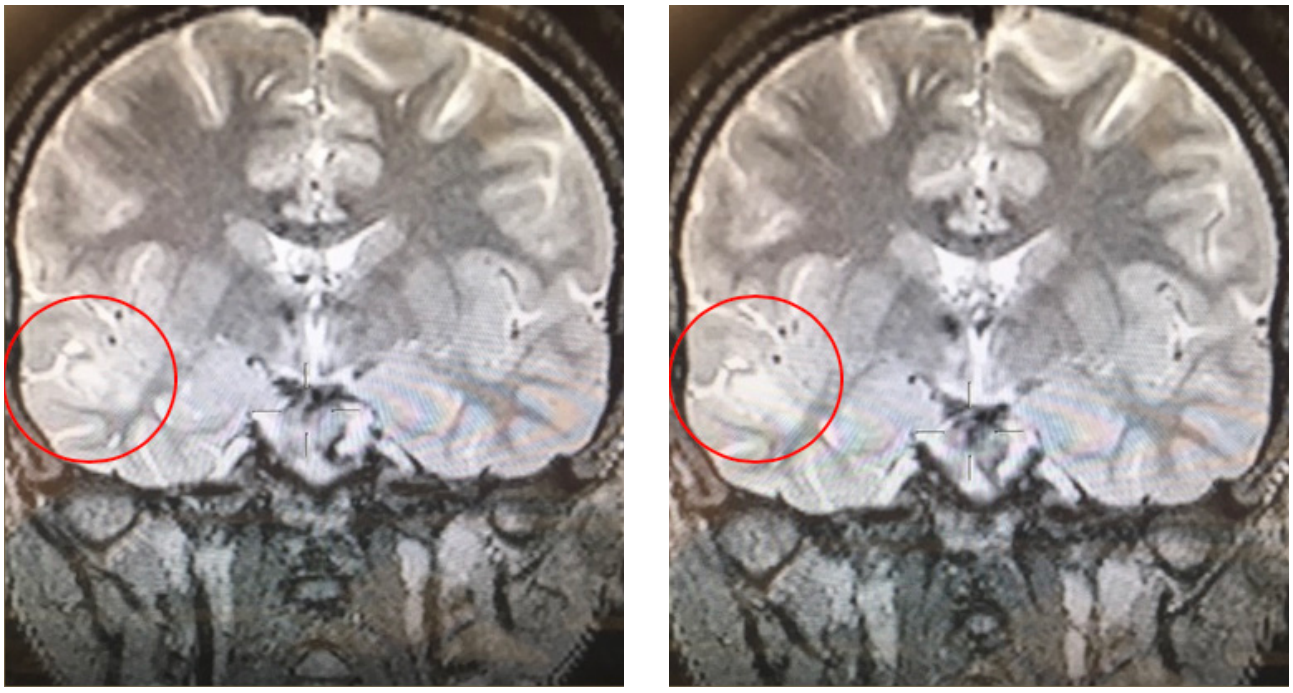

Figure 1. Hyperintense lesion in the superior temporal gyrus at the right temporal lobe on brain MRI

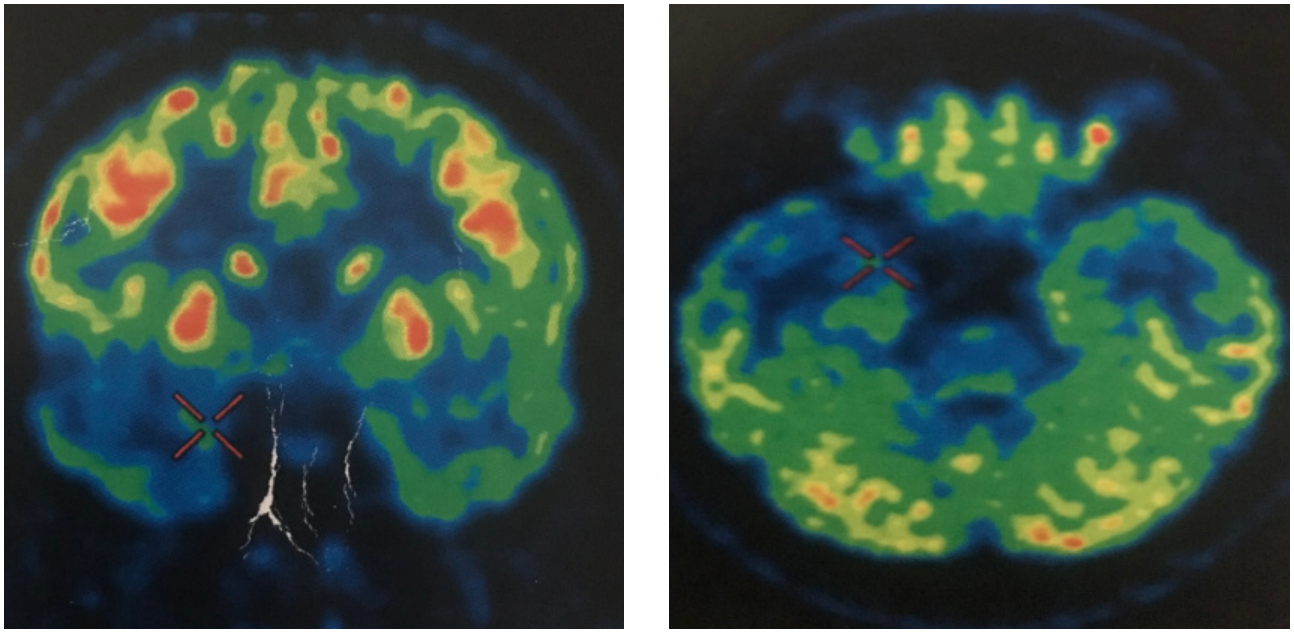

Figure 2. Focal hypometabolism in the right medial temporal cortex at the 'FDG-PET' images

ral region. The patient underwent video-EEG recording for 5 days, and two seizures were recorded after anticonvulsant drug reduction. The seizures were characterized by staring for a few seconds, during which the patient stayed unresponsive. This was followed by spitting and singing, the same song each time (Video 1). The song was from a television-drama series that he had watched during his childhood and had an emotional impact on him. Ictal EEG showed rhythmic theta-wave activity on the T4 standard electrode.

Brain magnetic resonance imaging detected a subcortically located hyperintense lesion in the superior temporal gyrus of the right temporal lobe (Figure 1). Fluorodeoxyglucose positron emission tomography imaging revealed focal hypometabolism in the right medial temporal cortex (Figure 2). As there was a hypometabolism in the medial temporal cortex and the lesion was located in the superior temporal gyrus, we decided to perform lesionectomy. Before the surgery, neuropsychological tests were performed, which re-

vealed defects of the executive functions and nonverbal memory, concordant with the nondominant temporal lobe damage. Histopathology demonstrated WHO Grade I ganglioglioma and cortical dysplasia type IIIb in the middle-inferior temporal gyrus.

After the surgery, his drugs were discontinued within 1 year, and he was seizure-free for 5 years without an anticonvulsant treatment till the last follow-up.

Written informed consent was obtained from patient who participated in this study.

Discussion

Automatisms are defined as coordinated and repetitive motor activities. During automatisms, cognition is impaired, and an amnestic period occurs [8]. A variety of automatisms with strong emotional components have been described, including sudden changes in facial expression seen with mimetic automatisms, uncontrollable laughter in gelastic seizures, and crying in dacrystic seizures [9, 10]. Ictal singing can be accepted as a rare form of automatism with an emotional connection. Although there is some knowledge about the underlying neuronal networks of normal singing, till date there is no information about the ictal singing mechanisms.

The auditory-motor integration for singing relies on the neural systems for vocalization. This auditory-vocal system comprises auditory, motor, and premotor regions in the dorsal stream as well as the cerebellum, basal ganglia, and brainstem structures [11, 12]. These also include the anterior insula, anterior cingulate cortex (ACC), and intraparietal sulcus. The previous studies on the neural correlation of vocalizations were performed in nonhuman primates that revealed the supplementary motor area (SMA)/pre-SMA and ACC to be important for initiating voluntary vocalizations [11, 12]. The regions of the brain associated with singing were thus introduced as the auditory-vocal network that regulates both behavioral features of vocal control in humans [13-15]. Looking at the subject in terms of lateralization, nonsignificant ictal vocalization has no lateralizing value, contrary to the meaningful speech with lateralizing value to the nondominant hemisphere [16].

Another question about the seizure semiology is which mechanism underlies behind singing the same song at each attack?

The underlying mechanisms of ictal automatisms are not well described. It is hypothesized that the automatisms may either be a reaction to an internal stimulus or the release of behavior by the removal of an inhibitory control [17]. Epileptic seizures cause excitation as well as the inhibition of neuronal activity, leading to behavioral dysfunction [18]. Regarding singing, the wide variance of the involved areas suggests that the mechanism of ictal singing is probably related to the recruitment of music-specific neural networks involving different regions in both the hemispheres of the brain, rather than the activation of a specific cortical region.

Our patient sang the same song during every attack. The song was from a television drama, which was based on a tragic love story, where a young man was compelled to marry his brother's widow because of the local village tradition, although he was in love with another girl. The patient had watched the series during his childhood and was emotionally influenced by it. Therefore, it might be the release of subconscious emotions suppressed during childhood, which stimulated him to recall the song and sing it during the seizures. Seizure semiology is usually discussed from a mechanistic point of view, considering every case to be similar, disregarding the personal history and differences of the patients. However, certain behaviors during the seizures may be interpreted from a different perspective, including the psychological background of each patient.

To conclude, singing during a seizure is a rare ictal phenomenon related to music-specific networks, which may be a reflection of a specific content associated with personal memories.

You can reach the video of this article at https://doi.org/10.5152/cjm.2020.20001.

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