Ictal singing due to left mesial temporal sclerosis

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ABSTRACT – Simple ictal or interictal automatisms are commonly seen in epilepsy, whereas complex automatisms are rare. Simple ictal vocalizations or appendicular automatisms have been reported to be due to mesial temporal lobe epilepsy. However, seizure-associated singing and dancing is very rare. We report a seventeen-year-old patient with ictal singing and rhythmic swinging of the arms as a dancing gesture. The video-EEG recording showed ictal left temporal lateralization and neuropathology confirmed left mesial temporal sclerosis. [Published with video sequences].

Key words: ictal singing, automatisms, epilepsy, temporal lobe

Case report

A seventeen-year-old, right-handed girl presenting with brief episodes of singing and rhythmic arm movements. She was the first child of healthy, non-consanguineous parents born by normal vaginal delivery. She had normal physical and mental development. Her uncle and grandfather had febrile convulsions and her mother’s aunt had seizures after the age of 40. She had suffered febrile convulsions when she was 20 months, two and a half years and four years old, for which she was treated with carbamazepine. After the age of four, she started having dialeptic seizures once every six to seven months under carbamazepine treatment. When she was ten, 1 000 mg/day sodium valproate was added to her treatment, and the dialeptic seizures decreased. The EEG performed at that time was normal, and the antiepileptic treatment was
Two weeks later, she had a generalized tonic clonic seizure (GTCS) and sodium valproate 750 mg/d was reintroduced. The EEG and CT were normal at that time.

She had experienced only four generalized seizures until she was 14, when she was hospitalized for agitation, crying spells, insomnia, and hallucinations. The EEG revealed bi-hemispheric, bioelectric disorganization. Cranial MRI showed left mesial temporal sclerosis (figures 1 and 2). This episode was diagnosed as interictal psychosis. She was given antipsychotic treatment and her symptoms resolved.

With 1 000 mg/d sodium valproate treatment she had rare seizures, however, she reported awakening from sleep with fear, “feeling hot” and described fearful visual hallucinations such as “horsemen approaching”. She had also meaningless speech for a short period during this time.

Two years later, she had another episode of interictal psychosis.

When she was seventeen, her mother described brief episodes of singing and swinging of both arms seen as primitive dancing, showing rhythmicity with the melody. Long-term video-EEG monitoring was performed and five seizures were recorded. The seizures started with fearful auras and feeling hot, followed, in two seizures, by singing and swinging of the arms, “freezing” in three, and immobilization of the right hand in two. Postictal nose-wiping with either hand, and anoma were recorded after two seizures.

The EEG recordings showed rhythmic theta activity in the anterior and middle temporal (F7, T1, T3) regions within 10 seconds after the clinical onset of the seizures. This activity partially spread to the left frontal region, ending up with post-ictal delta activity in the same regions. Frequent interictal spike and wave activity was also recorded in the left temporal region (figure 3; see the video sequence).

The clinical and electrophysiological findings were found consistent with left mesial temporal sclerosis. She underwent selective left amygdalohippocampectomy. Neuropathology revealed severe neuronal loss in CA1, moderate neuronal loss in CA2 and CA3, minimal neuronal damage at the dentate structure and partial neuronal loss of the amygdale showing hippocampal sclerosis.

She had over 90% right hand, feet and eye dominance according to the Edinburgh Handedness Inventory and Harris Test of Lateral Dominance. Before the operation, her neurocognitive examination revealed only a mild nonverbal memory deficit. At 12 months follow-up she had a mild deterioration in verbal memory, mild attention deficit but full recovery of nonverbal memory.

She has been seizure-free for 22 months with antiepileptic treatment.

**Discussion**

Ictal singing and dancing are very rare, complex automatisms, the localization remaining controversial.

Only five cases have been reported so far, of which there was only one with a right frontal lobe lesion which was accompanied by ictal, rhythmic slapping of the thighs as a dancing gesture.

Vidalhilet et al. (1989) reported a case of left frontal tumor with no EEG correlate. Ictal events varied in terms of melody and lyrics, which were usually related to her daily activities. Meierkord and Shorvon (1991) reported another case with a normal CT scan and non-lateralizing ictal EEG, whose seizures manifested with different words and melodies. The interictal EEG showed right temporal discharges.

Doherty et al. reported two more cases. Their first case referred to a patient with a pituitary tumor, who underwent two resections; the first employing a left temporal and the second a transphenoidal approach; this was complicated...
by bacterial meningitis. His EEG revealed multifocal, ictal discharges with dominance over the left frontotemporal region, with no ictal lateralization. The other patient had left mesial temporal sclerosis on MRI. However, the ictal discharges, including the ones with singing, were localized to the right temporal lobe as seen by invasive video-EEG monitoring (Doherty et al. 2002). The last case by McChesney-Atkins et al. (2003) with right frontal encephalomalacia due to a right transtemporal subfrontal resection of a craniopharyngioma, revealed ictal right frontal discharges. This patient displayed rhythmic slapping with both hands, although not always correlated with the rhythmicity of the melody (McChesney-Atkins et al. 2003).

Interestingly, our patient was very fond of music, singing songs most of the day and wanted to be a singer. Her desire decreased after the operation but she still likes to accompany singers. Of the other five cases, one of them was a choir director, another one came from a musical family, could play piano, trumpet and tuba and sang songs, another patient loved popular music and could sing a song after hearing it a couple of times. Although there is no clear evidence, there might be an association between the automatisms and personal interests.

Music production requires the involvement of different components such as rhythm, melody, lyrics, pitch, harmony, timbre and musical memory. It seems that complex neural networks play a role in the composition of these different components. Although there is probably no single cortical representation for musical ability, there is some evidence of association with the right hemisphere from automatisms of complex partial seizures, amobarbital injections, intraoperative cortical stimulation studies and case reports of musicogenic reflex epilepsy (Gordon and Bogen 1974, Penfield and Perot 1963, Wieser et al. 1997, Dennis and Hopyan 2001).

It was shown that the right superior temporal cortex participates in the perceptual analysis of melodies, whereas interaction of right temporal, frontal and prefrontal cortex plays a role in pitch comparison and retention (Zatorre et al. 1994).

A PET scan-based study showed that musical abilities such as tune recognition, pitch and rhythm lateralize to the left hemisphere, whereas timbre lateralizes to the right (Platel et al. 1997).

Samson and Zatorre (1991) reported that lyric recognition was impaired after left temporal resection, whereas tune recognition was impaired after right temporal resection. Similarly, it has been shown in several studies that left temporal lobe function was more closely associated with the verbal aspects of musical expression, whereas the right temporal lobe was involved with the nonverbal aspects (Samson and Zatorre 1992, Zatorre and Halperin 1993, Chauvel et al. 1998).

Interestingly, association of new lyrics with a familiar melody was impaired irrespective of the temporal lobe resected (Samson and Zatorre 1991). Our case is unique in terms of ictal singing and dancing gestures, showing rhythmicity with the melody, and with ictal left temporal lateralization.
References


