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Clinical letter

Does ictal whistling help to lateralise

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We present a right-handed, young female with a history of epilepsy since the age of 11. She also suffered from systemic lupus erythematosus, requiring treatment with hydoxychloroquine.

The patient described no warning signs that preceded her seizures. Her seizures were associated with loss of awareness for approximately 1 min followed by post-ictal confusion for 5 min. During some of her seizures, whistling was reported by witnesses. Her seizures clustered around her menstrual cycle. She was treated with different antiepileptic medications, such as carbamazepine, phenytoin, zonisamide, pregabalin, and oxcarbazepine. Her treatment regimen upon admission to the epilepsy monitoring unit included levetiracetam, phenobarbital, and lacosamide.

She was admitted for video EEG monitoring for a presurgical evaluation for refractory epilepsy. She experienced two to five seizures per month.

Video EEG monitoring with telemetry revealed the timing and symptomatology of her seizures.

During the five days of video EEG monitoring, two different types of seizures were recorded. Her seizures were associated with loss of awareness and whistling initiated in the left hemisphere, reaching a maximum onset in the left temporal lobe, specifically at the midtemporal region at T5 electrode. Her second type of seizures, associated with staring and unresponsiveness, were initiated in the right temporal lobe.

The left temporal lobe-onset seizures initiated as rhythmic sharp theta waves in the left temporal lobe approximately 10 s after the clinical onset of whistling. The video EEG revealed a spread of the field to the right hemisphere within 10 s, followed by diffuse delta waves. Towards the end of each seizure, delta frequencies exhibited greater amplitude and laterality in the left posterior quadrant. The total duration of the seizures with left temporal lobe onset was approximately 2 min. Clinically, the patient exhibited whistling followed by staring with a lack of awareness of her surroundings, followed by a gaze preference to the left with head deviation to the same side. She experienced restless movements of her left upper and lower extremities. Whistling was the first identifiable clinical sign observed during these seizures. Whistling was always initiated approximately 10 s before scalp EEG onset. There were no other clinical signs or automatisms recorded. The patient experienced post-ictal confusion for a few minutes afterwards (Figs. 1 and 2; Video 1: left temporal onset seizure).

The right temporal lobe-onset seizures initiated as rhythmic sharp theta discharges in the right temporal lobe, with a spread to the left hemisphere at times. Clinically, the patient paused her activities and became unresponsive, with staring but without any automatism. No other clinical symptoms were observed. These seizures lasted approximately 2 min (Figs. 3 and 4; Video 2: right temporal onset seizure).

The patient experienced a total of two right temporal lobe-onset seizures: one recorded on the first day of admission and the other on the second day. There were a total of five left temporal lobe-onset seizures with whistling, three of which were recorded on the second day and two of which were captured on the third day of hospitalisation.

The administration of all of the antiepileptic medications began to be tapered upon admission.

Interictal and epileptiform discharges and focal slowing were detected in the temporal lobes of both hemispheres, in addition to the left central and posterior quadrant regions. An MRI scan revealed several small foci of increased signal intensity in flair and T2-weighted images, primarily within the subcortical white matter tracts of both cerebral hemispheres.

1. Discussion

Whistling is a complex motor process that involves the coordinated articulation of the lips, the teeth, and the tongue. The process involves various cortical and subcortical areas of the brain. Complex neuronal networks linking the inferior rolandic cortex, the cingulate cortex, the basal ganglia, the amygdala, the thalamus, and the cerebellum have been shown to be involved in whistling behaviour, based on functional imaging.[1] The phenomenon of ictal whistling can be described as an automatic behaviour

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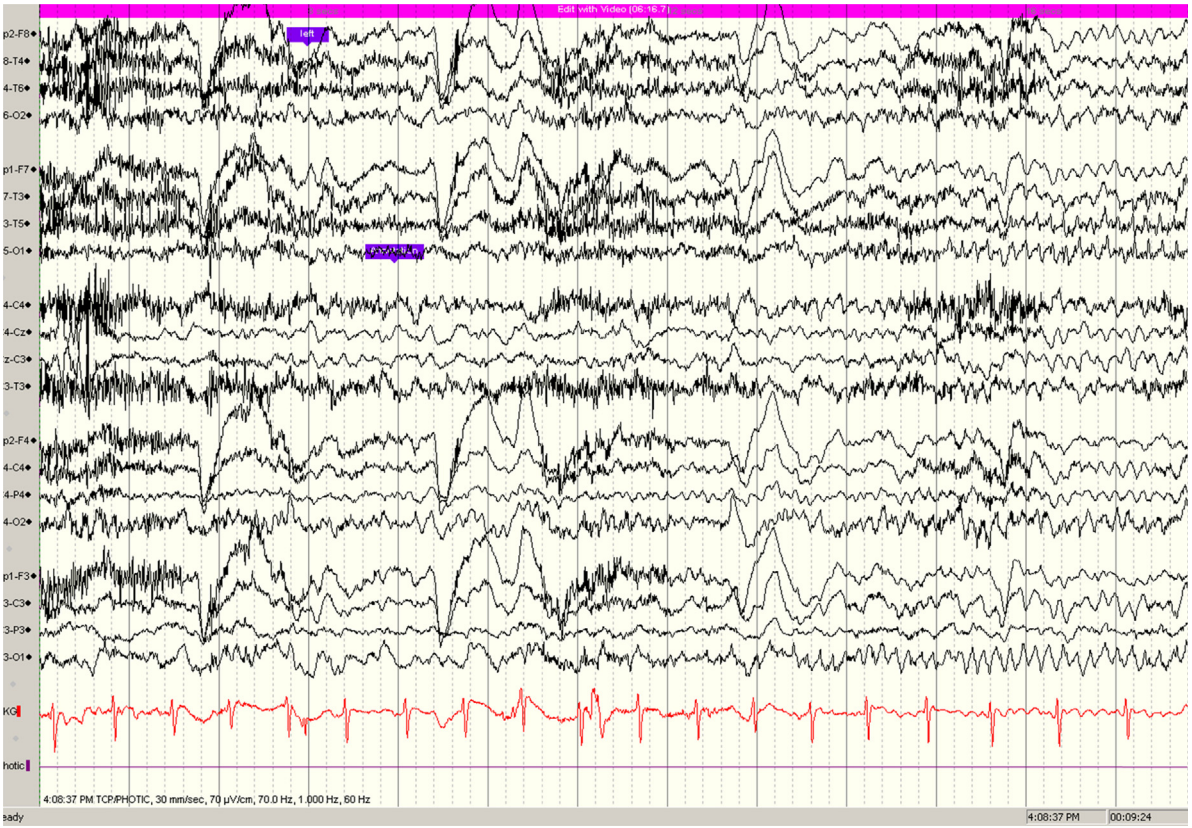

Fig. 1. Whistling seizure; clinical onset as marked by purple notations precedes EEG onset of rhythmic discharges over the left temporal area. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

Fig. 2. EEG onset with rhythmic theta discharges over left temporal area.