Localization of pilomotor seizure demonstrated by electroencephalography/functional magnetic resonance imaging

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Abstract

We report the first case of a pilomotor seizure detected by electroencephalography/functional magnetic resonance imaging (EEG/fMRI). An adult woman presented with history of bouts of gooseflesh feeling and pilomotor activity in the left leg following viral encephalitis. 24-hour video-EEG and simultaneous EEG during fMRI revealed ictal discharges in the right parietal and temporal lobes. Associated blood oxygen level-dependent (BOLD) activations were found mainly in the right parietal region. The result represents a different generator of pilomotor seizure compared to prior reports. We suggest that the feeling of gooseflesh could be the core ictal symptom and a direct pathway from the sensory cortex to the lower autonomic system may exist bypassing the classic cerebral autonomic center.

Key words: Autonomic, EEG/fMRI, pilomotor seizure

Introduction

Autonomic symptoms, such as cardiovascular, papillary, gastrointestinal, sudomotor, vasomotor, and thermoregulatory functions, are common during many types of epileptic seizures. As one of the associated signs of thermoregulation, pilomotor activity is not rare, especially in temporal lobe epilepsy. However, as the principal and independent ictal manifestation and subtype of autonomic seizure, pilomotor seizure is the least common. The origin of pilomotor seizure has been debated on, and most studies found that it can be detected through scalp EEG, intracranial EEG, magnetoencephalography (MEG), or single photon emission computed tomography (SPECT). Recent advancements in EEG/fMRI provide a more direct and new means to detect seizure origins. The demonstration of the origin of ictal pilomotor activity by EEG/fMRI has not been documented. We reported a patient of secondary unilateral pilomotor seizure with the ictal sign studied by EEG/fMRI and discussed the value of its localization.

Case Report

A right-handed 30-year-old woman was admitted in 2008 because of strong and frequent bouts of gooseflesh feeling and pilomotor activity in the extensor aspect of her left leg, forearm, or both. In the preceding months, she was hospitalized for fever, headache, and two attacks of general tonic-clonic seizure. Magnetic resonance imaging (MRI) was normal. Cerebrospinal fluid examination revealed a slightly increased nucleated cell count of 16×10⁹/L and normal protein and glucose. She was treated with anti-viral drugs. Three-dimensional and fast fluid-attenuated inversion recovery (FLAIR) MRI done at this admission showed no abnormal signals. However, 24-h video-EEG confirmed seconds-long seizures of strong goose bumps on her left leg, not accompanied by cold shiver, pallor, flushing, or sweating. Ictal discharges revealed focal slow waves on the right parietal and temporal lobes, but no epileptiform discharges or unilateral predominance were found during the interictal period. The spells
occurred once or twice per hour. Daily medication of carbamazepine (300 mg) controlled the seizures for 2 weeks, and then the dose had to be doubled to stop the seizures.

EEG was continuously recorded by the Galileo NT software through 21 MRI-compatible silver/silver chloride electrodes with 12 kΩ current-limiting resistors on the scalp according to the 10/20 system during fMRI scanning. The MR artifact was filtered offline with BE-MRI Toolbox (Galileo New Technology, Florence, Italy). Imaging was performed on a 3Tesla GE Horizon EchoSpeed system (EXCITE, Milwaukee, WI, USA). The patient was in the resting state with eyes closed. Data on seven successive runs of 6 min 40 sec each were acquired with EEG/fMRI (30 slices, 200 volumes, TE=30 ms; TR=2000 ms; thickness=5 mm, field of view=24 cm×24 cm; matrix = 64×64, flip angle=90°). The piloerection was marked according to the patient’s right hand gesture immediately after the attack and the descriptions of her and her husband accompanying in the scanner room.

fMRI data were analyzed using Statistical Parametric Mapping (SPM2) software package (http://www.fil.ion.ucl.ac.uk/spm). Blood oxygen level-dependent (BOLD) images were realigned, normalized, and spatially smoothed with a Gaussian kernel of 6 mm full-width-at-half-maximum. Maps of the T statistic were created with the onset of ictal discharge as the event in the analysis. Maps were obtained using four hemodynamic response functions (HRFs) peaking at 3, 5, 7 and 9 s after the onset of the seizure. The special activated areas were calculated using statistical t-tests with five contiguous voxels above a |t| value of 3.14 (P<0.001, uncorrected). For each voxel, the largest absolute T-value was adopted.

There were six runs acquired. Only one episode was captured during the scanning. Seconds of gooseflesh were felt on the extension part of the left leg and pilomotor activity was viewed by her husband. Simultaneous EEG revealed slow wave discharges in the central, parietal, and temporal regions with right predominance. The associated BOLD signals revealed that activations mainly lay in the right parietal region [Figure 1].

### Discussion

Pilomotor activity is common as an aura or accompanying symptom of seizure. It may occur either uni-or bilaterally and is often accompanied by other autonomic symptoms as chills, sweating, or pallor, and so on. It is usually considered as the manifestation of temperature dysregulation. Its origin is still under debate. Normal temperature regulation is controlled by the central autonomic network, which includes the hypothalamus, insula, potine parabranchial complex, periaqueductal grey matter, medial prefrontal cortex, nucleus of tractus solitaries, and ventrolateral medulla. The literature suggests that ictal piloerection is typically associated with seizures originating in the temporal lobe. Other seizure origins such as the frontal, fronto-parietal, fronto-temporal, or parieto-occipital regions have also been reported.

All the above reports are analyses by scalp EEG. To our best knowledge, this is the first report detecting the origin of pilomotor seizure with EEG/fMRI which can reveal the more direct and precise temporal distinction.
in demonstration of focal blood flow and oxygen metabolism. The patient had pilomotor activity as the independent and single sign of the seizure, and the strong feeling of gooseflesh was the predominant subjective symptom for her to notice the seizure aside from usual focal or systemic autonomic symptoms such as sweating, flushing, and others. The fMRI showed the associated activation mainly in the contralateral postcentral gyrus, otherwise, the typical temporal lobe and central limbic structures did not show any activation. This result presents an evidence of the different generator of pilomotor activity, as compared to prior reports.\textsuperscript{[1,2,6-9]} We therefore suggest that the feeling of gooseflesh is the core ictal symptom, while piloerection is the result of the feeling. Other than pilomotor seizure, focal sensory seizure may be a better description for our patient. On the other hand, the result shows a direct pathway in charge of piloerection from the sensory center to the lower autonomic center. Autonomic regulations bypassing the classic autonomic center are totally possible. Though, in most cases, they are involuntary. What we have presented in this work is just one special case. Further research is certainly needed for a clearer understanding of the precise neural pathway of pilomotor activity.

References


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