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Letter to the Editor

Micturition and startle-induced reflex seizures in a patient with focal cortical dysplasia in the middle frontal gyrus

Reflex seizures arise from hyperexcitable cortex within the areas physiologically activated during specific sensory stimulations, and cognitive or motor activities. Micturition-induced seizures are rare, only a few clinical cases have been reported, all with normal neuroimaging findings, and EEG indicative of fronto-central onset (Whitney and Callen, 2013; Jang et al., 2018). Startle seizures are induced by sudden and unexpected stimuli, usually noise. Most of the patients have widespread structural brain abnormalities, predominantly in the frontal, temporal and perisylvian cortices (Palmini et al., 2005; Yang et al., 2010). Here we describe a patient with both micturition and startle-induced reflex seizures due to focal cortical dysplasia in middle frontal gyrus.

A 21-year-old man was initially seen in our center several years ago with recurrent seizures from the age of 9 years. He had had spontaneous seizures for some years before startle sensitivity developed. His seizures occurred consistently, immediately after an unexpected noise, but not if the same stimulus was anticipated. Micturition-induced seizures supervened lately and the event triggering these seizures was passing urine. The seizures occurred a few times daily. Bowel opening did not trigger the episodes. He had a normal neurologic examination and IQ of 75.

We recorded numerous typical spontaneous and reflex seizures during three separate video-EEG monitoring sessions. The semiology of the majority of seizures consisted of brief axial and left arm tonic posturing followed by left extremities proximal movements. Frequently he will urinate during the seizure. Seizures during sleep reproduced the same pattern but usually followed by hypermotor component. Some of the spontaneous and micturition seizures consisted of loss of body tone and fall, with the eyes and head deviated to the left. Interictal activity consisted of focal rhythmic epileptiform discharges with the maximum over F4/Fp2/Fz electrodes, which become more continuous during drowsiness and non–REM sleep (Fig. 1A). Ictal EEG revealed fast-spike discharges over the right lateral frontal region (F 4 electrode), and almost simultaneously involving contralateral homologous regions and vertex. This was followed by low voltage fast activity which abruptly became generalized (Fig. 1B). The seizures persisted for 10–20 s and were followed by bilateral frontal slowing. Magnetic resonance imaging (MRI) revealed focal cortical dysplasia in right middle frontal gyrus (Fig. 1C).

Our patient is an uncommon example of coexistent micturition and startle elicited seizures. Additionally, to our knowledge, this is the first case of micturition-induced seizures with MRI confirmed structural lesion.

Patients suffering from startle seizures usually have structural abnormalities resulting from prenatal or perinatal insult and involving large areas of sensorimotor cortices. However, stereo-EEG studies demonstrated a constant involvement of the lateral and mesial premotor cortex in seizure generation, thus exhibiting the pivotal role of dorsolateral premotor and supplementary motor (SMA) cortices. It is still a matter of debate whether an unexpected stimulus triggers both the startle and the seizure, or whether it elicits first a normal startle reaction that in turn precipitates the seizure.

Micturition-induced seizures are rare and poorly understood, and all previously reported cases exhibited normal MRI findings. However, the majority have been reported to have an EEG focus observed near midline (Cz) or in the right medial frontal region (F 4). Based on these findings the possible mechanism of micturition-induced seizures was proposed: while voiding, midline (SMA and anterior cingulate) and frontotemporal regions that play a role in micturition might be excessively activated and seizures are generated shortly after the voiding process (Whitney and Callen, 2013; Jang et al., 2018). Several possible types of stimuli were implied to trigger a seizure during urination, such as somatosensory and viscerosensory. In our patient, seizures occurred soon after starting urination and we deduce that actually proprioceptive stimuli from detrusor activation may play the crucial role. Available evidence indicates that F 2 (premotor) neurons respond not only to somatosensory stimuli but in particular to the proprioceptive ones. Urodynamic studies can also be helpful to understand how micturition induces seizures as one study reported phasic detrusor overactivity with a normal urinary flow (Seth et al., 2014).

It is interesting that different types of stimuli led to reflex seizures, suggesting that both stimuli projected to the dorsolateral premotor cortex, where an FCD was found. Subsequently, hyperexcitable dorsolateral premotor cortex (which primary function is an integration of sensory and motor information for the performance of an action) seems to play an essential role in the generation of micturition and startle seizures in our patient. Seizure semiology reflects the involvement of larger networks in lateral and mesial premotor cortices, anterior cingulate, and a probably negative motor area in drop attacks. Previous reports included variable motor components such as tonic posturing and clonic movements for micturition seizures (Whitney and Callen, 2013; Jang et al., 2018), while startle-induced were usually described as generalized tonic, myoclonic, asymmetric tonic and atonic seizures (Palmini et al., 2005; Yang et al., 2010). Finally, of previously reported cases of startle as well as micturition-induced seizures, many patients had a developmental delay. Our patient also has a borderline deficiency. The plausible explanation would be that the rostral premotor areas, located in between motor and prefrontal cortices, seem to function as a “gateway” hooking up both motor and cognitive

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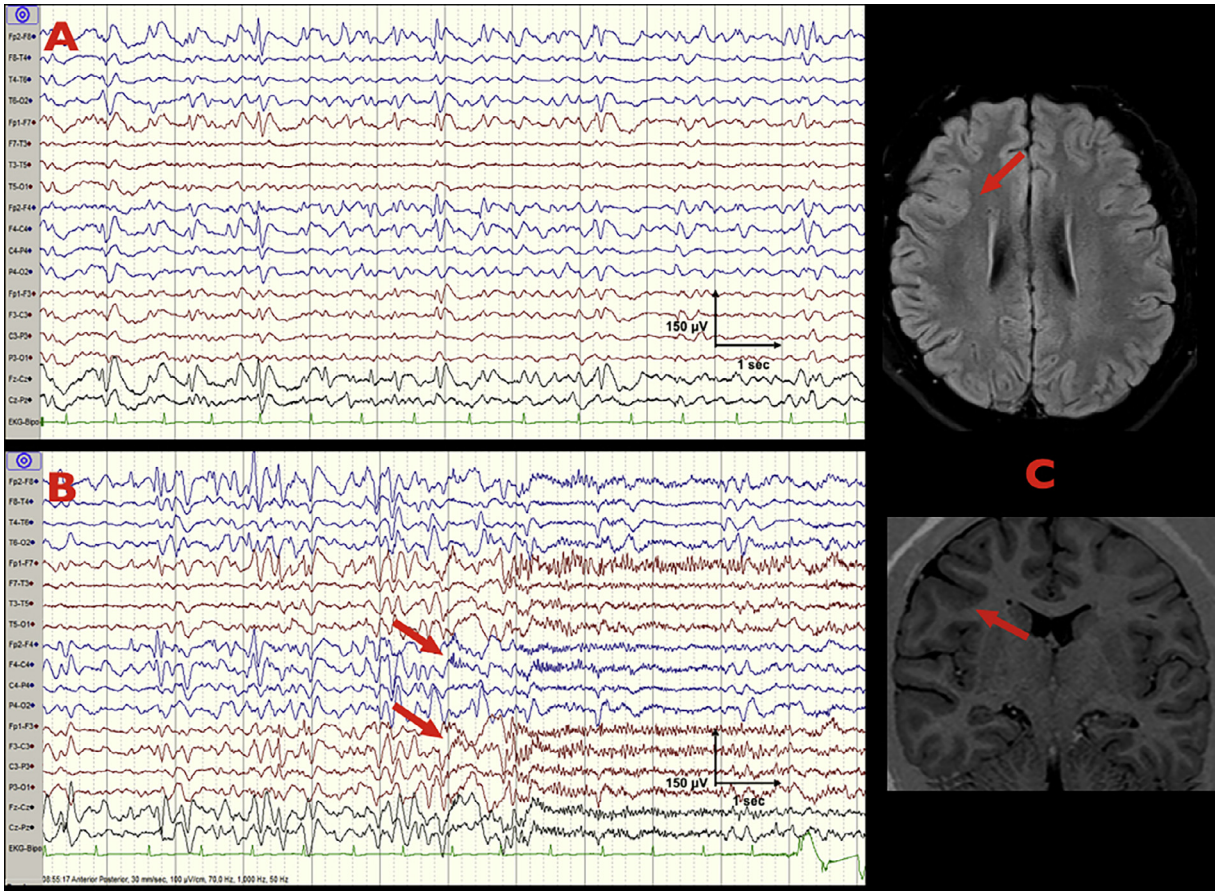

Fig. 1. Reflex seizures in a patient with FCD in middle frontal gyrus. A: frequent interictal epileptiform discharges with a maximum over F4/Fp2/Fz electrodes. B: Ictal EEG during reflex startle seizure (onset is indicated by red arrows). Fast-spike discharges over the right lateral frontal region (Max F 4 electrode), and almost simultaneously involving contralateral homologous region and vertex, followed by low voltage fast activity which abruptly became generalized. C: FLAIR and T1-weighted MRI show focal cortical dysplasia in the right middle frontal gyrus.

networks, i.e. the site where the cognitive and motor networks interact.

Conflict of interest

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