



## Ictal dancing-like semiology in frontal lobe epilepsy

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Dear Editor,

Dancing is a very rare seizure semiology and it is characterized by an integrated gestural motor behavior of rhythmic swinging of the arms and pelvis resembling a dance. Seven cases presenting with dancing-like ictal/post-ictal behavior have been previously reported, but the underlying mechanisms and neural networks of this complex and fascinating ictal behavior remain still not completely understood [1–4]. The epileptogenic zone (EZ) has been previously hypothesized to be localized both in the temporal and in the frontal lobes. Some authors suggested that the dancing semiology seems to be not typical of temporal lobe epilepsy, since automatisms involving proximal muscles and complex behaviors, such as bicycling, are suggestive of frontal lobe epilepsy [1, 4, 5].

We report a case of a 33-year-old right-handed woman who suffered from medically refractory, weekly, or daily focal seizures, in which the clinical semiology was marked by sudden onset of repetitive dancing-like behavior that lasted a few seconds. Loss of contact was inconstantly reported during seizures. According to the report of her husband, movements would have sometimes assumed a form of a twirling dance, making a right turn when she was standing. No family history or risk factors for epilepsy have been reported. She was admitted to our center to explore the feasibility of epilepsy surgery. Brain MRI showed a possible hyperintensity of signal in T2

FLAIR-weighted sequences, located in the parasagittal aspect of the right frontal lobe (Fig. 1a).

Interictal awake and sleep EEG showed focal epileptiform abnormalities on the right frontal and central contacts, as well as right hemispheric spike-and-wave discharges. Her habitual seizures were recorded during video-EEG monitoring and were clinically characterized by sudden fixed facial expression, mild dystonic posturing of the left arm, and rhythmical movements involving the arms and both legs, similar to stepping through a dance, as well as repetitive rhythmical movement involving the trunk and the pelvis, like a “twist” dance (see additional file: [video](#)). No post-ictal signs were observed except for mild pelvic movements in the bed. She did not report any preceding aura and was completely amnesic with respect to the episodes. Ictal EEG showed a biphasic slow wave on the right fronto-central and temporal regions with superimposed recruiting low-voltage fast activity followed by muscular artifacts related to the movements. Neuropsychological evaluation revealed a visual long-term memory deficit. An invasive study by using subdural electrodes was offered to better define EZ. The strategy of electrode positioning was defined after the revision of presurgical data in a multidisciplinary case conference. After craniotomy under general anesthesia, three 4-contact strips were positioned over the right frontal medial cortex, while two 6- and 8-contact strips were positioned in the dorsolateral right frontal cortex, centered on the supposed lesion. Finally, one 10-contact depth electrode was inserted orthogonally in the left frontal lobe to control the contralateral medial cortex (Fig. 1b). Two habitual focal seizures were recorded, with ictal EEG correlate visible on contacts investigating the posterior dorsolateral parasagittal right frontal cortex, rapidly spreading on the ipsilateral posterior medial frontal cortex and then on all contacts. Interictally, subcontinuous epileptiform discharges were also recorded in the same contacts where seizures started (Fig. 2a, b).

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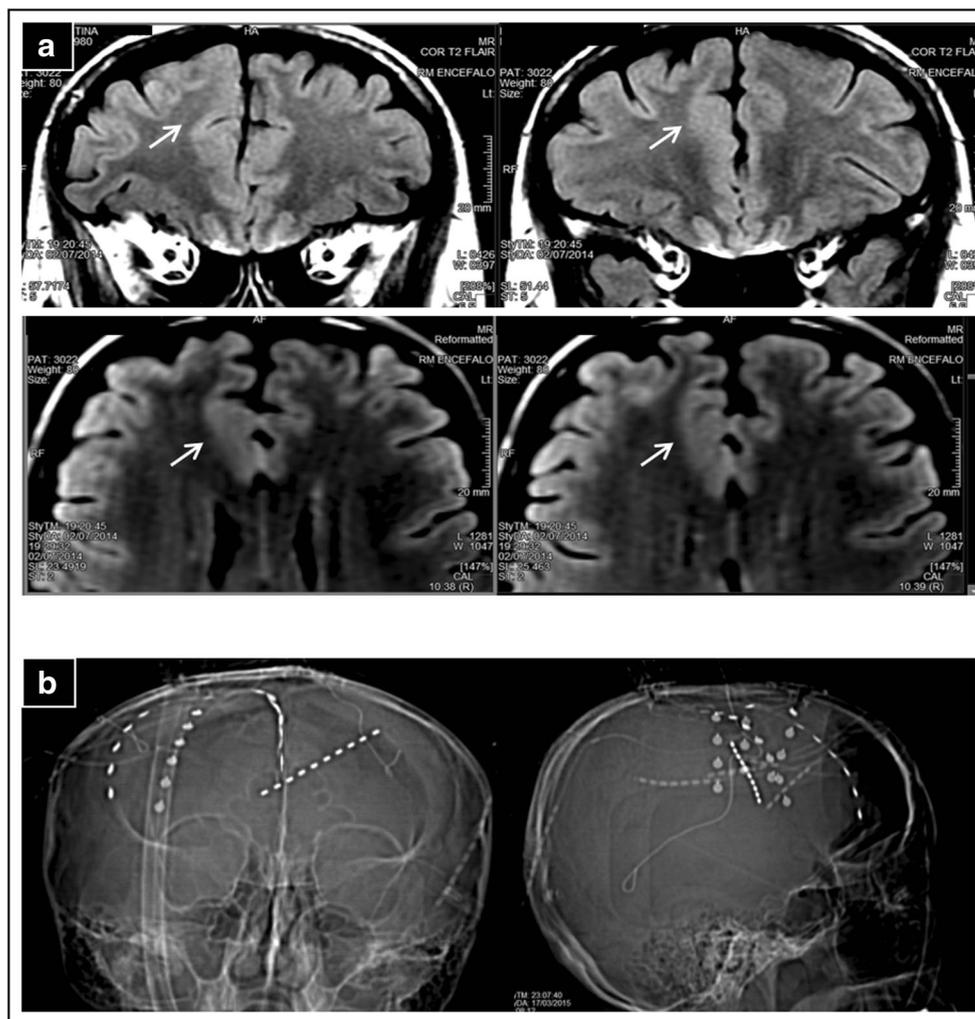
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**Fig. 1** **a** Brain MRI (coronal and axial T2 FLAIR-weighted sequences) showing a possible hyperintensity of signal located in the parasagittal aspect of the right frontal lobe. **b** Coronal and sagittal X-ray scan showing three 4-contact strips positioned over the right frontal medial cortex, two 6- and 8-contact strips positioned in the dorsolateral right frontal cortex, centered on the supposed lesion and one 10-contact depth electrode inserted orthogonally in the left frontal lobe



Based on invasive investigation findings, she underwent frontal right cortectomy, with evidence of heterotopic neurons in white matter at histopathological examination. At her last follow-up, the patient had been seizure-free for 24 months, thus beginning a slow discontinuation of antiepileptic medications.

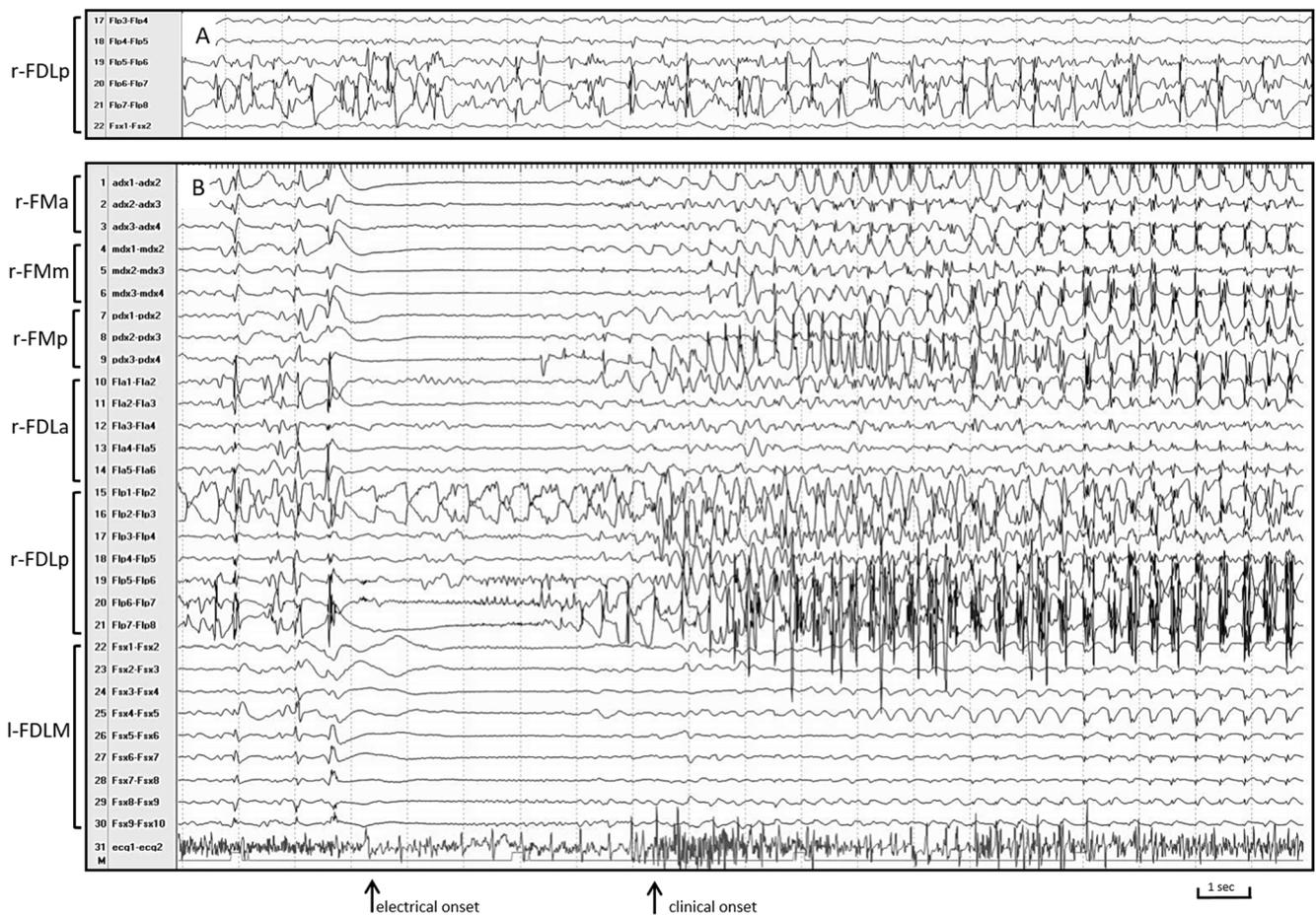
The case we reported describes a clinical semiology rarely encountered during focal epilepsy, consisting in an integrated gestural motor behavior composed of repetitive and rhythmic movements mainly involving the trunk and the extremities, resembling a dance. Although such a hyperkinetic clinical semiology evokes seizures originating in frontal lobe structures, dancing seizures were observed both in temporal and in frontal lobe epilepsy, thus suggesting a wide epileptogenic network involving both structures [1–5].

In previous reported cases, resective surgery was performed in two subjects affected by temporal lobe epilepsy due to hippocampal sclerosis (TLE-HS) (anterior temporal lobectomy and selective amygdalohippocampotomy, respectively) and in one patient with symptomatic FLE (lesionectomy), allowing a sustained seizure freedom in all cases [2]. One patient underwent stereotactic gamma knife

surgery applied to the atrophic hippocampus and complained one seizure at a 3-month follow-up visit [3].

For all, except one [3], previously reported cases, clinical, MRI, and neurophysiological finding and surgery and outcome data were reviewed in detail by Limotai and Noivong [2].

Although achieving seizure freedom after surgery is essentially the confirmation of EZ complete removal, this condition does not allow conclusions to be drawn regarding the cerebral substrate of semiologic features, since the distributed brain networks involved in producing ictal signs will in many cases involve structures distant from the zone of seizure onset. In this context, invasive EEG investigations, including seizure recording, may consistently shed more light on this complex issue. In fact, Bartolomei and colleagues [4] reported a patient with ictal singing associated with dancing who was investigated by using stereo-encephalography (SEEG), showing intracranial ictal EEG changes involving the right dorsolateral and medial prefrontal regions. Recently, other authors [5] investigated ictal motor patterns in subjects with FLE by using SEEG and found that an ictal integrated behavior tends to occur in anterior aspects of the frontal lobe. In accordance with these findings, also in our



**Fig. 2** a Interictal intracranial EEG pattern showing subcontinuous epileptiform discharges over contacts investigating the posterior dorsolateral parasagittal right frontal cortex. **b** Ictal EEG correlate of habitual focal seizure recorded during invasive EEG study, visible on contacts investigating the posterior dorsolateral parasagittal right frontal

cortex, rapidly spreading on the ipsilateral posterior medial frontal cortex and then on all contacts. r-FDLp, right frontal dorso-lateral posterior; r-FMa, right-frontal medial anterior; r-FMm, right-frontal medial medial; r-FMp, right-frontal medial posterior; r-FDLa, right-frontal dorso-lateral anterior; l-FDLM, left-frontal dorso-lateral-medial

patient, who was investigated by using subdural electrodes, the EZ was hypothesized to involve the antero-medial portion of the frontal lobe, thus supporting the hypothesis that these regions are involved in the networks underlying this complex gestural motor behavior [5]. Theoretically, the connectivity of frontal lobe areas supports spread through distant multilobar and multidirectional cortico-cortical pathways, commonly resulting in rapid and widespread propagation of ictal discharges originating in frontal regions, thus helping to explain the semiologic clinical complexity.

In conclusion, to date, the underlying neural networks of ictal dancing remain still not fully understood. Further cases also studied by using intracranial investigations are needed to elucidate neural substrates and networks of this unique and fascinating semiology.

### Compliance with ethical standards

**Conflict of interest** The authors declare that they have no conflict of interest.

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