Clinical commentary

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Ictal kissing in a patient with right frontal lobe epilepsy

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ABSTRACT – When performing pre-surgical evaluation of patients with refractory epilepsy, the analysis of seizure semiology is one of the key elements used to generate a hypothesis about the location of the epileptogenic zone. Ictal kissing is a very rarely observed ictal automatism described in patients with temporal lobe epilepsy. We present a 62-year-old man who was referred to our epilepsy centre for comprehensive evaluation. During prolonged video-EEG monitoring, six focal-onset hyperkinetic seizures were registered. In five seizures, the patient repeatedly produced sonorous kisses "*into the air*". Initial ictal EEG pattern consisted of rhythmic theta or alpha activity at the right fronto-polar and fronto-medial electrodes. MRI depicted focal cortical dysplasia located in the right prefrontal medial cortex. This case suggests that ictal kissing can also occur in the setting of right frontal lobe epilepsy; we therefore believe that this observation expands the anatomo-clinical correlation for this rare ictal automatism. [*Published with video sequences*].

Key words: ictal kissing, frontal lobe epilepsy, seizure semiology

When performing pre-surgical evaluation of patients with refractory epilepsy, the analysis of seizure semiology is one of the key elements used to generate a hypothesis about the location of the epileptogenic zone. Some clinical features are common and well known to have high lateralizing and/or localizing value (clonic limb movement, dystonic hand posture or post-ictal aphasia, to name a few examples) (Foldvary-Schaefer and Unnwongse, 2011). Other symptoms and signs are rarer and their anatomical networks are incompletely understood. Ictal kissing fits this last category as it is observed very rarely and its localizing and lateralizing value are hard to establish. Ictal kissing has previously been reported in patients with definite or presumed temporal lobe epilepsy (Alsemari *et al*., 2013; Özkara *et al*., 2016; Asadi-Pooya and Fletman, 2017). We present a patient in whom ictal kissing was a consistent feature of ictal semiology and all available data pointed towards a right frontal lobe epileptogenic zone.

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Case study

A 62-year-old man with a diagnosis of epilepsy since age 17 was referred to our refractory epilepsy centre for comprehensive evaluation. The patient and family reported three types of paroxysmal events:
– auras with a feeling of discomfort and anxiety;
– agitation and incoherent speech with variable loss of awareness;
– and generalized convulsions.
Seizures with agitation occurred once a week on average and generalized convulsions had not been occurring for many years. He had no known epilepsy risk factors. The only co-morbidity was dyslipidaemia. At the time of admission, he was prescribed pregabalin (150 mg bid), controlled-release carbamazepine (400 mg bid), topiramate (50 mg bid), and clobazam (20 mg id). He had previously failed other antiepileptic drug (AED) trials, including levetiracetam and lacosamide. There was no family history of epilepsy. General and neurological examination were unremarkable. Magnetic resonance imaging (MRI) showed an area of thickened grey matter and blurred grey-white matter transition in the right prefrontal medial cortex (anterior cingulate), highly suggestive of focal cortical dysplasia (*figure 1*); mesial temporal structures were normal. Neuropsychological evaluation revealed executive dysfunction and preserved episodic memory.

Video-EEG monitoring was performed over five days, with AED reduction. Six focal-onset hyperkinetic seizures, some with awareness and some with impaired consciousness, were recorded, all from N1 or early N2 sleep, with a mean duration of 70 seconds (46-95 seconds). Semiology was characterized by a change in facial expression suggesting disgust (with sustained downward movement of labial commissures, *i.e.* pouting), hyperkinetic pelvic and bilateral proximal limb movements, and vocalizations and one-word nonsensical, sometimes obscene, verbalizations. Early during the evolution of five out of the six recorded seizures, he repeatedly produced sonorous kisses "into the air", *i.e.* unaccompanied by attempts to reach bystanders (*video sequences 1 and 2*) This ictal behaviour occurred early during seizure evolution, either during or outside interaction with the technicians who performed ictal testing (mean duration from seizure onset was 11 seconds). Some lateralizing features were noted since he had dystonic posture of the left arm in four seizures and consciousness was preserved in two seizures. He had no post-ictal confusion. The initial ictal EEG pattern consisted of rhythmic theta or alpha activity at the right fronto-polar and fronto-medial electrodes (also involving right anterior temporal electrodes, but with significantly lower amplitude), later spreading to contralateral fronto-temporal regions (*figure 2*). Interictally, occasional multifocal epileptiform activity on the bilateral fronto-polar, right anterior temporal

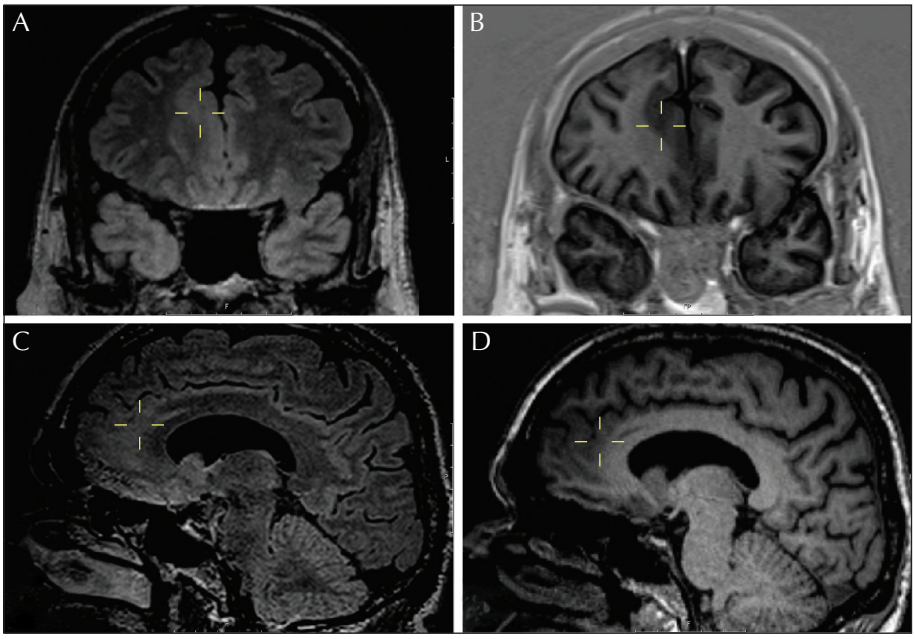

Figure 1. MRI sequences: (A) coronal FLAIR; (B) coronal T1; (C) sagittal FLAIR; (D) sagittal T1, showing thickened cortex and blurring of grey-white matter junction in the right anterior cingulate region, suggesting focal cortical dysplasia.

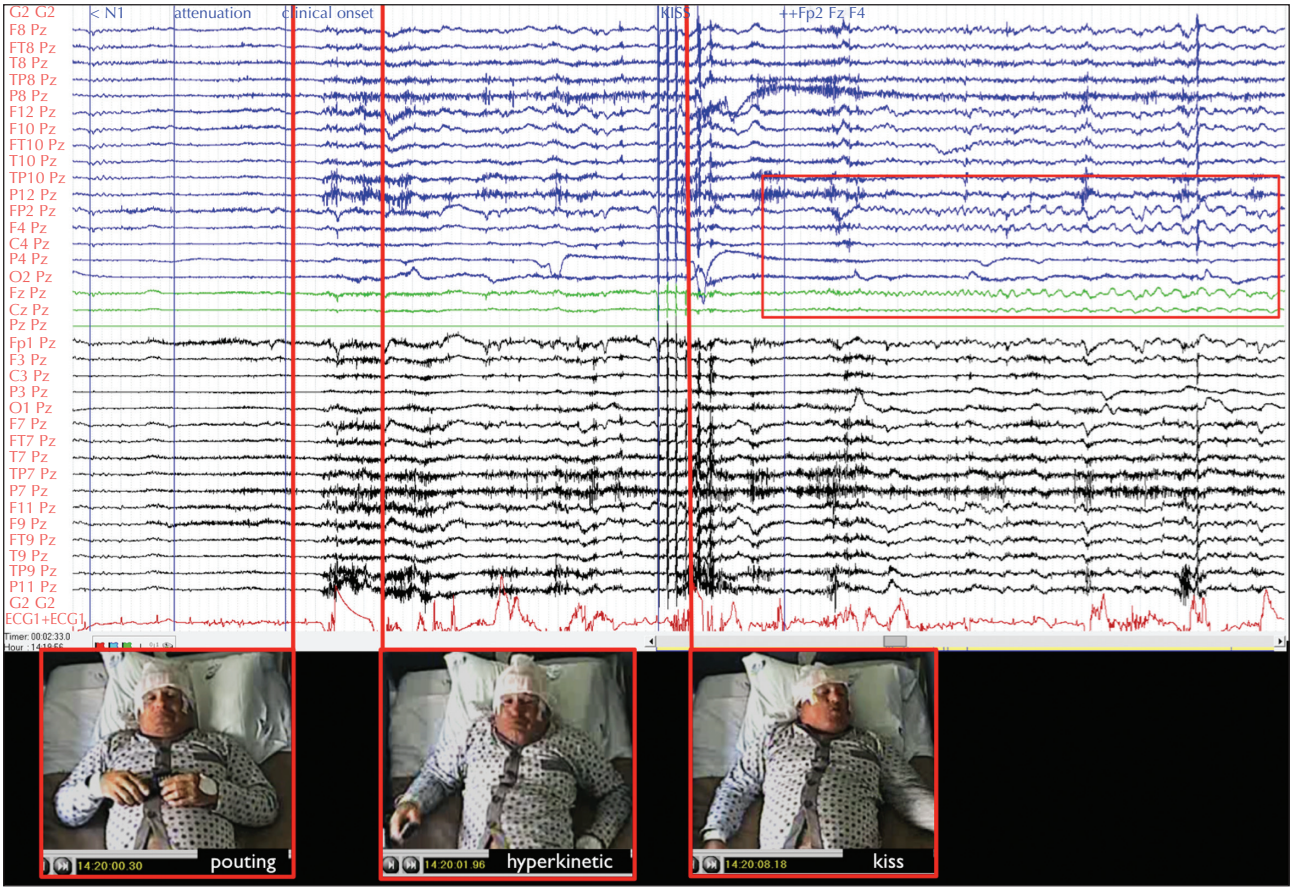

Figure 2. EEG and snapshots of semiological features: pouting, axial hyperkinetic movements, and kissing. A regional right frontal ictal pattern (alpha, then theta sinusoidal rhythm) is seen, 8 seconds after clinical onset. (reference=Pz; notch off, high-frequency filtre=70 Hz; low-frequency filtre = 0.53 Hz; reference gain=300 uV/cm; 25 seconds/page).

and bilateral fronto-temporal regions were recorded. After discharge, AED adjustment was made and the patient has had a sustained reduction of seizure frequency. The patient declined epilepsy surgery.

Discussion

An automatism is defined as a more or less coordinated motor activity, usually occurring when cognition is impaired and for which the subject is usually amnesic afterwards (Fisher *et al.*, 2017). Automatisms may be seen in focal seizures and in absence seizures. Ictal kissing is an automatism very rarely observed during focal seizures (Alsemari *et al.*, 2013; Özkara *et al.*, 2016). In a cohort of 5,133 patients, the prevalence of ictal kissing was 0.02%, corresponding to a single patient (Alsemari *et al.*, 2013). At our centre, we have performed video-EEG monitoring on 350 adult patients from 2012 to 2018, and the patient presented is the only one who presented with this semiological feature. To the best of our knowledge, there are 26 cases reported in the literature (Alsemari *et al.*, 2013; Özkara *et al.*, 2016). The kissing pattern may not be stereotypical among seizures in the same patient (Özkara *et al.*, 2016). Brain MRI revealed lesions in 19 cases: within the temporal lobe in 17, consisting of mesial temporal sclerosis in 15 (12 on the right side); within the right parietal lobe in one; and in the right frontal lobe in another. Predominance of hippocampal sclerosis in these case series may reflect a selection bias as all patients reported were evaluated at epilepsy monitoring units. The epileptogenic zone was considered to be on the right temporal lobe in 17 cases and on the left temporal lobe in four cases; in five cases, the localization was unclear due to normal or bilateral MRI and EEG findings. The patient with the parietal lesion had ictal discharges propagating to the right temporal lobe at an early phase (Özkara *et al.*, 2016). In the case reported by Alsemari *et al.*, the MRI showed "right frontal small cortical and subcortical lesions", but eventually subdural EEG recording,

covering the right frontal and temporal lobes, showed that seizures started at the right mesial temporal lobe contacts. In summary, in all published data, the finding of ictal kissing has been consistently associated with temporal lobe epilepsy. According to Jasper (1964), automatisms may result from several mechanisms: a reaction to internal stimuli; an activation of a cortical-specific motor programme or behaviour; or a result of an inhibitory effect leading to a behaviour-release phenomenon. Since cortical stimulation has never elicited kissing behaviour (Selimbeyoglu and Parvizi, 2010), it is highly unlikely that this mechanism is responsible for ictal kissing and so no specific epileptogenic zone can be accurately related to this automatism. In addition, as mentioned earlier, kissing behaviour is not stereotypical among seizures in the same patient. This might reflect the ability to respond to the surrounding environment and react to external cues (*e.g.* blowing kisses towards a technician who has asked the patient to follow a command, or trying to kiss a nearby family member) or the activation of more widespread network connections with bilateral temporal lobe involvement and resemblance of a Klüver-Bucy syndrome with hyperorality and a more sexualized kissing behaviour (Özkara *et al.*, 2016; Rashid *et al.*, 2010). It has also been proposed that a faster spread of ictal discharges to both temporal lobes might be responsible for a self-initiated kissing behaviour (*e.g.* blowing kisses into the air) (Rashid *et al.*, 2010). However, we should keep in mind that published case reports are based on scalp EEG recordings which do not capture the involvement of deep structures and do not provide completely reliable real-time spreading patterns. For this reason, the exact network involved cannot be determined. Interestingly, in the only case report using deep electrodes (Alsemari *et al.*, 2013), kissing behaviour was correlated with the spread of epileptic discharges to the frontal lobe, allowing us to speculate that the frontal lobe may have an important role in ictal kissing.

The semiology of frontal lobe seizures includes, depending on anatomical localization and seizure spread patterns: elementary motor signs such as clonic movements and tonic posturing; gestural motor behaviour (hyperkinetic movements such as pedalling or movements with attempts to fight or escape); non-verbal vocalizations, screaming or swearing; and negative or positive facial emotional expressions, among others (Bonini *et al*, 2014). According to the work of Bonini *et al*. (2014), patients with frontal lobe seizures manifesting negative facial expressions, swearing, and hyperkinetic movements, without elementary motor signs, typically have seizures involving the medial-prefrontal region, propagating to anterior temporal regions, which is in line with our patient's ictal EEG pattern. In our patient, kissing behaviour occurred after or during hyperkinetic activity, in contrast with published literature in which ictal kissing followed typical oral and manual automatisms associated with temporal lobe seizures. Since it has been shown that the frontal lobe is important for kissing behaviour (Alsemari *et al.*, 2013), we propose that when kissing emerges in the context of typical frontal lobe ictal signs, it should be considered as part of a frontal lobe seizure.

Since our patient declined epilepsy surgery, the location of the epileptogenic zone was not confirmed. However, multiple convergent data clearly support a diagnosis of right frontal lobe epilepsy: a right frontal lesion suggestive of focal cortical dysplasia, semiology (hyperkinetic seizures arising from sleep [Alqadi *et al.*, 2016] and ictal pouting [Souirti *et al.*, 2014]), initial ictal EEG pattern (clearly maximum in the right frontal region), and distribution of interictal epileptiform activity (which included fronto-polar spikes). We are convinced, therefore, that in our patient, ictal kissing occurred within the setting of right frontal lobe epilepsy and believe that this case expands the anatomo-clinical correlation for this rare ictal automatism. □

Legend for video sequences

Representative examples of the patients' focal hyperkinetic seizures. Pouting is seen initially, then agitation with predominantly axial movements, at which time the kissing behaviour is seen. Awareness is not entirely lost, as some degree of interaction is seen during parts of the evolution of the seizure. Post-ictal recovery is fast and complete. For synchronous EEG (Pz referential montage) for both seizures, after an initial stage of diffuse attenuation, an evolving ictal pattern is seen involving initially the right frontal derivations, very close to the timing of the kissing automatisms.

Key words for video research on www.epilepticdisorders.com

Phenomenology: ictal kissing
Localization: right frontal lobe
Syndrome: frontal lobe epilepsy
Aetiology: focal cortical dysplasia

Supplementary data.
Summary didactic slides are available on the www.epilepticdisorders.com website.

Disclosures.
None of the authors have any conflict of interest to declare.

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TEST YOURSELF

(1) Ictal kissing is a rare ictal automatism usually associated with:
A. Frontal lobe epilepsy
B. Temporal lobe epilepsy
C. Occipital lobe epilepsy

(2) Frontal lobe seizure semiology does not include which of the following:
A. Clonic movements and tonic posturing
B. Complex motor behaviour
C. Complex visual hallucinations

Note: Reading the manuscript provides an answer to all questions. Correct answers may be accessed on the website, www.epilepticdisorders.com, under the section "The EpiCentre".