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## Ictal kissing: occurrence and etiology.

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## **Brief communication**

**Title:** Ictal kissing: occurrence and etiology

**Running title:** Ictal kissing

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## **Abstract**

Ictal kissing is a rare semiological manifestation in patients with epilepsy. We tried to estimate its occurrence and characterize the underlying etiology. We retrospectively reviewed all video-EEG reports from Jefferson Comprehensive Epilepsy Center over a 12-year period (2004-2015) for the occurrence of the term “kiss”. We then searched the electronic database PubMed on September 21, 2016 using the following search terms in the English language: “ictal” OR “seizure” OR “epilepsy” AND “kiss” OR “kissing”. Relevant original studies were included. During the study period, 5133 patients were investigated in **our epilepsy monitoring unit**. One patient (0.02%) had one episode of documented ictal kissing. He had drug-resistant right mesial temporal lobe epilepsy. In total, 5 studies met the criteria for inclusion in the review. These studies described nine patients with drug-resistant focal epilepsy and ictal kissing behavior. Six out of 10 patients with ictal kissing had hippocampal sclerosis. We add to the literature on ictal kissing by providing additional information on its frequency and etiology. The most common underlying etiology for ictal kissing is hippocampal sclerosis. However, this semiological ictal phenomenon is not pathognomonic for any etiology or localization.

**Key words:** Epilepsy; Ictal; Kissing; Seizure

## **1. Introduction**

Seizure semiology is a simple and efficient tool that allows localization of the symptomatogenic zone, which often overlaps or is in close proximity of the epileptogenic zone. This becomes particularly important in patients with drug-resistant epilepsy who are potential candidates for surgery. Many semiological findings have been well described in the literature; some are valuable in localizing or lateralizing the epileptogenic zone [1]. Ictal kissing is a rare semiological manifestation in patients with epilepsy [2-6]. In this study, we describe a patient with ictal kissing to add to the literature on this rare phenomenon. We also review the literature to depict a clear picture of occurrence and etiology of ictal kissing.

## **2. Material and Methods**

We retrospectively reviewed all the long-term video-EEG reports from Jefferson Comprehensive Epilepsy Center over a 12-year period (2004-2015) for the occurrence of the term “kiss” in the text body. We defined “kiss” as a touch or gesture with the lips as a sign of love, sexual desire, or greeting (to be differentiated from lip smacking, which is a common automatic behavior in patients with epilepsy). All the extracted reports were reviewed and patients with at least one episode of documented ictal kissing in epilepsy monitoring unit (EMU) were included in the study. For each included patient, the data were reviewed from the electronic medical records, EMU report, and neuroimaging records. This study was conducted with approval by Thomas Jefferson University Institutional Review Board.

We then searched the electronic database PubMed on September 21, 2016 using the following search terms in the English language: “ictal” OR “seizure” OR “epilepsy” AND “kiss” OR “kissing”. Relevant original studies were included.

### 3. Results

During the study period, 5133 patients were investigated in our EMU. One patient (0.02%) had at least one episode of documented ictal kissing. He was a 63-year-old left-handed man with a history of seizures starting at age 30 years. He had drug-resistant complex partial seizures with an aura of epigastric sensation with a frequency of 1-2 per month. His interictal EEG showed right anterior temporal spikes and the ictal EEG showed right anterior temporal seizures. **During the long-term video-EEG monitoring**, he had four seizures, all happened while he was asleep. He woke up, had mouth automatisms and had laughing and giggling. During one of the seizures, he said “oh baby” and threw kisses at the nurse **(as a gesture of love or sexual desire)**. He was not responsive and did not remember the code word. Postictally, he was confused and did not move his left extremities for a few minutes. In brain MRI, right hippocampus was slightly small with no signal change. The PET scan showed right temporal hypometabolism. Neuropsychological tests showed impairment of visual-spatial memory. He had a right anterior temporal lobectomy in 2011. Pathology showed reactive gliosis in amygdala and hippocampus. He had a postoperative tonic-clonic seizure, 11 months after his surgery and then was lost on follow-up.

In total, 5 studies met the criteria for inclusion. Table 1 shows the characteristics of all the patients with ictal kissing including ours and those from the literature. **All the patients underwent long-term video-EEG monitoring.**

#### 4. Discussion

Automatisms are defined as more or less coordinated, repetitive motor activities during epileptic seizures, usually occurring when cognition is impaired and for which the patient is usually amnesic afterwards [7]. A variety of automatisms with strong emotional elements, such as sudden uncontrollable laughter in gelastic seizures [8] and crying in dacrystic seizures [9] have been well-described in the literature. Ictal kissing is a rare finding among patients evaluated in EMUs. The frequency of ictal kissing at our center was 0.02% and the total number of patients reported in the literature (including ours) is 10 [2-6]. So far, this rare semiological feature has just been reported among patients with drug-resistant focal epilepsy. However, no prospective study has ever looked for this semiological manifestation in patients with epilepsy. The patients who have been reported do not represent the full spectrum of people affected with epilepsy.

The most common underlying etiology for ictal kissing is hippocampal sclerosis, often on the right side. However, this rare semiological feature of focal epileptic seizures is not pathognomonic for any etiology or localization (Table 1). **However, the high proportion of hippocampal sclerosis among patients with ictal "kissing" may reflect a selection bias towards this etiology in epilepsy monitoring units.** Neuroimaging plays an important role in identifying the underlying etiology [10]. There is one patient with a right frontal cortical lesion (gliosis) [4] and another with a right temporal tumor [6] in the literature. In a case report of ictal kissing with subdural EEG recording, the seizures started in right mesial temporal electrodes, but the emotional kissing behavior was correlated with the spread of the epileptic discharges in the right frontal lobe [4]. This is an important consideration while dealing with patients who are potential candidates for epilepsy surgery. Patients with ictal kissing behavior during their seizures are

usually refractory to medical therapy; they may or may not respond well to resective brain surgery with respect to seizure control.

This rare semiological phenomenon may be considered as a temporal lobe automatism, not restricted to temporal lobe, but mainly observed in patients with temporal lobe epilepsy. Ictal kissing automatism is very complex in nature and most likely involves various brain networks. It probably occurs as a release phenomenon rather than a cortical inhibition or stimulation [2, 5]. In addition, family dynamics, personality, lifestyle, and instincts may all play roles in the emergence of this rare automatism during an epileptic seizure [2].

In conclusion, we add to the literature on ictal kissing by providing additional information on its frequency and etiology. Ictal kissing is a rare finding among patients evaluated in EMUs. **The most commonly identified underlying etiology is non-dominant hippocampal sclerosis; however, it is not a specific or pathognomonic sign for any etiology or localization and this may reflect a selection bias towards this etiology in epilepsy monitoring units.**

## **5. Acknowledgments**

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