

Ictal kissing and religious speech in a patient with right temporal lobe epilepsy

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ABSTRACT – We report the case of a 25-year-old female patient with intractable complex partial seizures characterized by repetition of certain religious statements and a rather compulsive kissing behavior. Presurgical evaluation revealed a right-sided, mesial temporal focus and hippocampal sclerosis on MRI. After selective amygdalohippocampectomy, she has only occasional auras. Her rare and peculiar ictal manifestations are discussed in the context of semiology and pathogenesis. [Published with videosequences]

KEY WORDS: complex partial seizure, kissing, religious speech, ictal automatism, temporal lobe epilepsy, hippocampal sclerosis

Mesial temporal lobe epilepsy with hippocampal sclerosis (MTLE-HS) is the most common type of medically intractable epilepsy, which is known to be remediable by surgery. The seizure semiology may differ from one case to another in relation to the propagation of the ictal discharge and the site of the focus. Isolated auras and complex partial seizures (CPS) with different types of automatisms are common features. Although several types of automatisms have been described during CPS originating from different regions, to our knowledge, kissing as an ictal event has not been reported in the literature before.

Case report

A 25-year-old, right-handed female was delivered normally at term, after an uneventful pregnancy. Her history of epilepsy started with status epilep-

ticus at age seven days related to dehydration. She experienced a second seizure at the age of three and a half years, as a left hemiconvulsion and was treated with phenobarbital (PB). Five years later, phenobarbital was withdrawn as she had no seizure recurrence.

At age nine, she had sun-stroke followed by an episode of status epilepticus during the same night. Therefore, phenobarbital was initiated again and was continued for four years. After an eight-year seizure-free period, epigastric sensations and feelings of discomfort started at the age of 16, which lasted for one year and was then followed by complex partial seizures. She had two to three seizures in a month, which were aggravated during menstruation. On admission, she was on 500 mg/day topiramate, 1400 mg/day of carbamazepine and of 2400 mg/day vigabatrin.



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Interictal EEG revealed bilateral temporal spike activity, very prominent on the right side, with a propagation to posterior temporal regions of the same hemisphere. Cerebral MRI demonstrated right-sided hippocampal sclerosis (figure 1), and neuropsychological evaluation showed nonverbal memory impairment. Neurological and psychiatric examinations were normal.

Video-EEG monitoring revealed ictal discharge starting from the right sphenoidal and anterior temporal leads (figure 2a). There were four ictal events recorded. Seizure semiology usually started with a feeling of gastric discomfort, followed by repetition of a certain religious statement in Arabic "La eela'hay eel'allah, Muhammed'een resul'allah" (God is unique and Muhammed is his prophet) with a latency of seven to 25 seconds (mean 19.2 seconds), from the onset of the ictal discharge. Five-6 Hz high amplitude, semi-rhythmic theta activity was seen on frontal and temporal electrodes at EEG during this verbal automatism (figure 2b). This was followed by brief oroalimentary automatisms and a compulsive kissing behavior with a latency of 35-50 seconds (mean 40 seconds). She kissed everything in proximity, especially her right hand or asked for the hands of other people around her to kiss, and performed this action rather vigorously. She was always amnesic to the event and regained immediately. However, according to the information given by the parents, if her fiancé was around, she kissed him on the lips; if her mother was close, she kissed her on the cheek. During EEG recordings, she kissed our technician's hand

and at this moment, the ictal discharge was still ongoing on the EEG, with prominent bifrontal 2-3 Hz sharp countered slow waves and diffuse dysrhythmic activity (figure 2c). The dystonic posture of her left arm, observed concomitantly with kissing during the second seizure and just after kissing in the third recorded seizure, suggests that the kissing behavior was part of the ictal event. Data obtained from our noninvasive evaluation protocol enabled the decision for epilepsy surgery to be taken. She underwent selective amygdalohippocampectomy (SAH) successfully. Pathological examination revealed severe hippocampal sclerosis accompanied by microdysgenesis. She has been having occasional auras for 15 months since surgery, especially during her menstrual periods and her medication was reduced gradually to topiramate 200 mg/day and carbamazepine 600 mg/day.

Discussion

The ictal phenomena of TLE can be divided into broad categories such as motor, sensory, autonomic, experiential, emotional, cognitive, and psychiatric. Motor symptoms include automatisms such as lip smacking, straightening the hair, circular running, darting eye movements, nose wiping, staring, and twitching or jerking of upper or lower extremities. Emotional experiences include embarrassment; sadness and sudden crying; explosive laughter (gelastic epilepsy) usually without the feeling of happiness; peacefulness with a sense of serenity or of "being at one with the universe"; and, most commonly, fear. Experiential, emotional, and autonomic manifestations are most likely to be associated with lateralization of the seizure foci to the nondominant right hemisphere [1]. Here we describe a patient with right TLE who had seizures with a rich repertoire of ictal symptoms including motor, religious and emotional features.

During temporal lobe seizures, ictal speech manifestations can frequently occur although identifiable speech is not common. At this stage, loss of consciousness usually occurs, but sometimes the patient may remain responsive, even in conjunction with automatisms. Verbal automatisms may be repetitive or non-repetitive but when present, an ictal or immediate postictal, clearly intelligible speech is highly suggestive of a non-dominant hemisphere involvement [2]. The underlying mechanism was suggested by Serafetidinides and Falconer [3] to be either the release of the dominant hemisphere from inhibition by the nondominant hemisphere, or the overexcitation of the nondominant hemisphere. Prominent involvement of the nondominant right TL during ictal speech in our patient could be explained by both hypotheses. However, interestingly, the content of her speech had a religious theme and always occurred just before the kissing automatisms. "La eela'hay eel'allah, Muhammed'een resul'allah" (God is unique and Muhammed is his prophet) was the Arabic

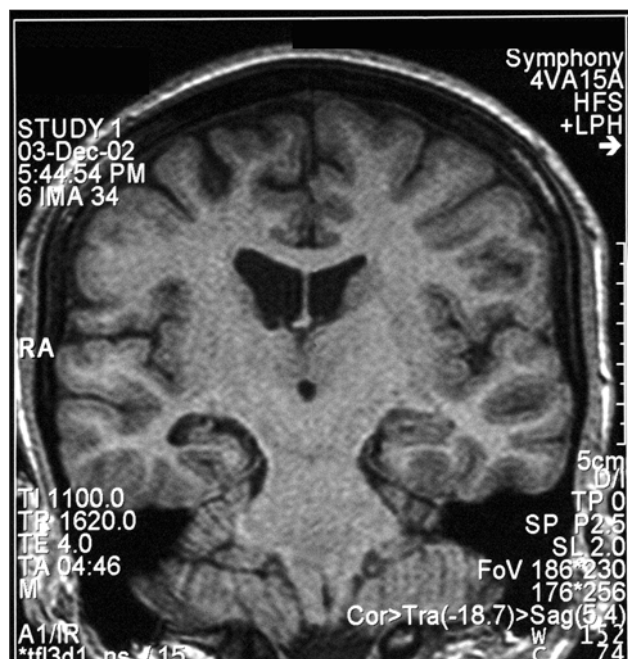


Figure 1. Right hippocampal sclerosis on MRI.

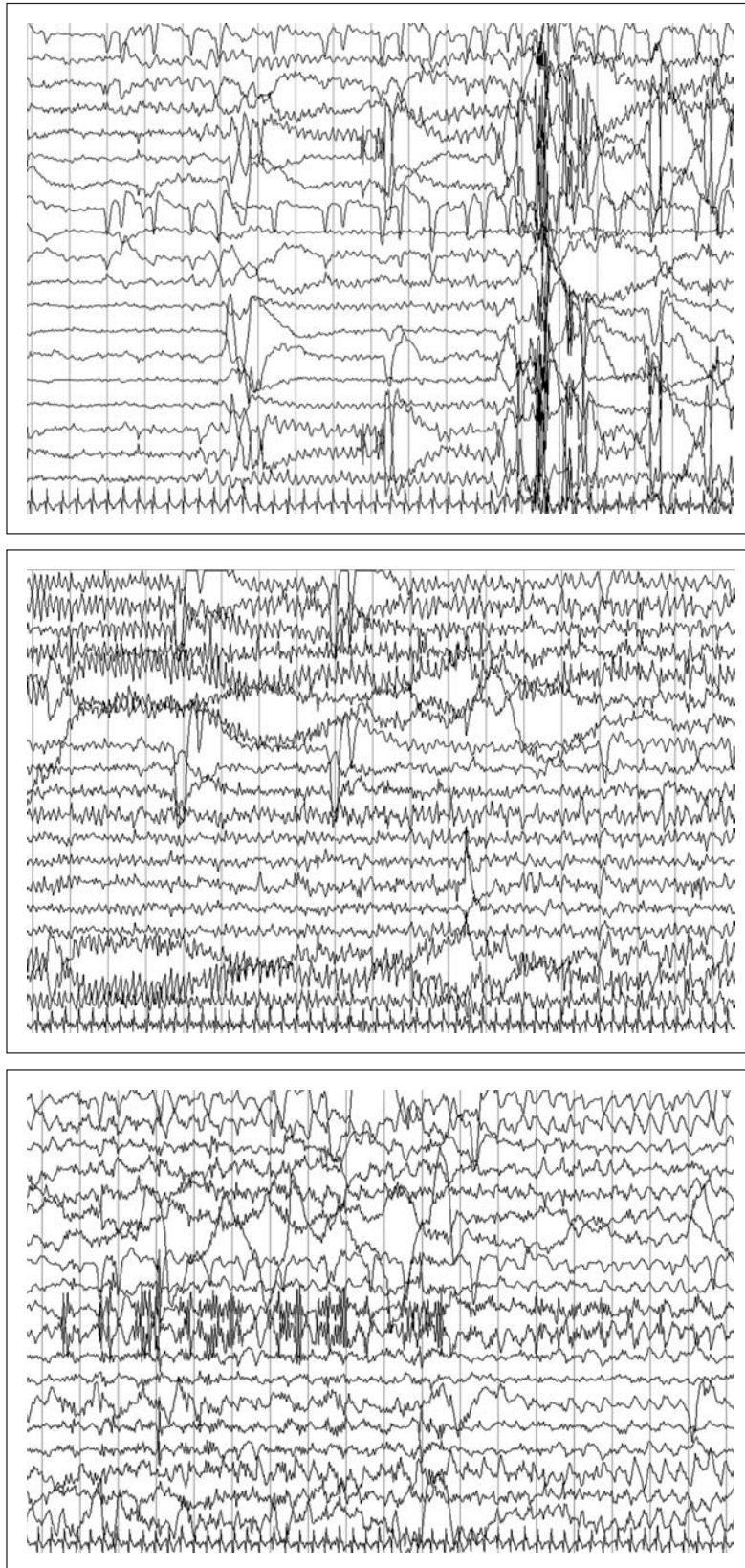


Figure 2. Ictal EEG: a) at onset of seizure; b) during ictal speech; c) during kissing automatism.

sentence spoken in a repetitive manner and sometimes with dysprosodia.

Ictal religious experiences have been found to be frequently associated with interictal hyperreligiosity and religious conversion. In one report [4], spiritual and religious experiential phenomena were reported in five patients, but no particular religious interests were present in four. Dewhurst and Beard described another six patients with temporal lobe epilepsy and sudden religious conversion experiences. In five of their cases, the conversion appeared to occur shortly after an isolated seizure or a cluster of seizures [5]. Our patient was a Muslim believer, however, she neither practiced religious rituals nor stated any extremely religious interests in her daily life. Therefore, religiosity was not the main characteristic feature of her personality, however, the contents of emotions liberated unconsciously during a seizure are unpredictable.

The kissing behavior was another interesting feature of her seizure. The act of kissing involved sexual motives in some of her seizures or turned out to be a compulsive behavior, with the repetition of kisses.

In the second seizure, she spat and blinked her eyes after kissing. During seizures two and three [video] she spoke, kissed and spoke again or there was intermixed speaking and kissing. She kissed the sheet only once in the fourth-seizure. There was dystonic posture during or immediately after kissing in the second or third seizure. The simultaneous EEG always revealed ongoing bilateral ictal activity predominant at the right and frontal leads. The other ictal phenomena and simultaneous EEG recordings confirmed kissing to be an ictal feature. To our knowledge, kissing was mentioned in only one case report about a nine-year-old boy whose seizures usually began 5-20 seconds after repeated cheek-kissing of his mother or after viewing pleasant or funny events. Non-emotional stimuli or the sound of the kiss alone failed to provoke seizures in this case [6]. Apparently, this patient had reflex epilepsy triggered by kissing, which is a different situation.

Nevertheless, kissing was thought to be an automatism in our case. In the Dictionary of Epilepsy, automatisms are described as "more or less coordinated, adapted (eupractic or dyspractic), involuntary motor activity occurring during the state of clouding of consciousness either in the course of, or after an epileptic seizure, and usually followed by amnesia for the event" [7]. Penry and Dreifuss divided automatisms into *de novo* and perseverative types. Perseverative automatisms are behaviors initiated prior to the ictus and continued during the ictus. *De novo* automatisms begin with or after onset of ictus and occur only during ictus. They are further classified as reactive, released (actions of normally socially inhibited behavior) and focal automatisms (conditional, unverified) [8]. Reactive automatisms represent the individual's ability to respond to stimuli (apparent or occult), when some of the neuronal pathways of the brain are not integrating infor-

mation properly and are not recording behavior in the memory. In the second seizure, our patient read the brand name of the cracker in her hand and repeated this word several times illustrating a reactive automatism. According to this classification, the kissing behavior may be interpreted in both ways: it can be a reactive behavior as the target of the kiss can be modified according to the "victim" (i.e. fiancé was kissed on the lips, the technician was kissed on his hand) or a release phenomena of a certain action which was manifested in relation to a social disinhibition. Klüver-Bucy syndrome is a good example of released behaviors reported in patients with bilateral temporal lobe lesions related to *Herpes encephalitis*, trauma etc. These patients lacked the ability to evaluate the affective significance of objects in their environment until they explored them orally [9]. However, there are only few case reports describing such phenomena related to epileptic seizures, and other symptoms such as hyperorality, hypersexuality, hypermetamorphosis and visual agnosia are usually associated. Therefore, this ictal behavior can not be easily explained within the full context of this syndrome. Nevertheless, release of the activity of nonaffected (unknown) regions during the seizure may still be considered by way of further explanation.

Jasper and Jackson suggested that automatisms might be the result of ictal activation of specific brain structures. They correlated them with electrical afterdischarges in the amygdala showing that automatisms result only with the spread of discharge to the hippocampus and involvement of lateral cortex and subcortical structures [10]. Moreover, a recent study which explored the electro-clinical correlations of humming during TL seizures showed that specific networks can be synchronized transiently during such an automatism, and that this synchrony may bind distinct functional areas remote to the epileptogenic zone [11]. Therefore, we can speculate about such networks operating emotional behaviors activated during kissing, which is a complex behavior in its nature, rather than accepting the presence of a kissing center in the brain. Furthermore, the close temporal relationship between religious verbal automatism and the kissing behavior deserves more intensive analysis in this patient, particularly if we take into consideration cultural and religious particularities.

Eye blinking or fluttering is a relatively uncommon ictal symptom. Although usually symmetric, unilateral blinking has been reported to be ipsilateral to the seizure focus in both extratemporal and temporal cases [12]. Our patient had bilateral eye blinking, prominent on the right side, and was ipsilateral to the focus in two of her seizures. Another uncommon ictal symptom observed was ictal spitting. It was observed during the second seizure, in between two episodes of kissing and can be included in the list of lateralizing signs for the nondominant temporal lobe. It was demonstrated to be the nondominant lobe when the association was with the left hemisphere [13].

Although our patient had several uncommon features occurring during her seizures, hand automatisms, which are known to be frequent in TLE and tend to be ipsilateral to the seizure focus mainly when associated with a contralateral dystonic posturing, were also present. During the seizures, she always preferred her right hand for automatisms, suggesting that this could also be a lateralizing sign.

Conclusion

Our patient presented with what we considered to be an ictal manifestation, characterized by a kissing behavior preceded by verbal vocalizations with a religious content. Whether these manifestations are the result of a release phenomenon or of activation of specific sites in the brain remains unclear and needs to be investigated further. □

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Legends for video sequences

1st seizure

She started with a feeling of discomfort; after 10 seconds, she started to repeat " *La eela'hay eel'allah, Muhammed'een resul'allah* " (God is unique and Mohammed is his prophet)" for twenty three seconds between 0:10-0:33. Then she said, "Give me your hand" and "My sweetie", and kissed the hand of technician using her right hand for 34 seconds. At 1:07, she again started to repeat " *La ilahe illallah, Muhammedin Resullulah* " for 14 seconds, followed by left hand automatisms (1:21).

2nd seizure

She started with a feeling of discomfort or fear (3: 14), and continued to grasp an object (biscuit) with both hands (3:25). After five seconds, she repeated " *La eela'hay eel'allah, Muhammed'een resul'allah* " (3: 29), and started to blow (4:02), she stretched her right arm toward the technician (4:23), kissed the hand of technician (between 4:27 and 5:05) said "Hold" and "My sweetie" (4:32), with her left arm in dystonic posture. At 5:07, she spat and this was followed by orolimentary automatisms. At 7: 00, the seizure stopped and at 7:05 she said that she felt good.

3rd seizure

She started with a feeling of discomfort or fear (7:21) At 7:39, " *La eela'hay eel'allah, Muhammed'een resul'allah* "

Left arm in dystonic posture, looking towards the left, stretching her right arm (8:04), saying "Hold" (8:11), starting to kiss the hand and said "Don't pity me" (8:18).

She blew and said, "Come" using two hands (8:57). In between, she repeated " *La eela'hay eel'allah, Muhammed'een resul'allah* " and blinked her eyes continuously. At 9:51, she continued to kiss. At 10:10, her left arm in dystonic posture and eyes deviated to the left side. At 11:20, she answered questions, and said that she felt better.

4th seizure

She started with a feeling of discomfort or fear (11:55). 12:18, " *La eela'hay eel'allah, Muhammed'een resul'allah* ", blinked her eyes continuously, her left arm was in dystonic posture.

13:01, she held the sheet and kissed it, then orolimentary automatisms started.

14:09, she put her right hand on her head.