



Ictal kissing: Electroclinical features of an unusual ictal phenomenon[☆]



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ABSTRACT

Purpose: The study aimed to describe the electroclinical features of ictal kissing, an unusual behavior that may occur during focal seizures.

Method: Twenty-five patients collected from four epilepsy centers and previously published reports were reviewed for their demographic, clinical, and electrophysiological features.

Results: Sixteen of 25 patients were female. The mean age was 32.9 years (9.9–51 years) and the average age at seizure onset was 14.5 years. All seizures were localized to the temporal lobe (TL) and lateralized to right side in 17 patients, left side in 2 patients, and unclear in 6 patients. A total of 55 ictal video electroencephalograms (EEGs) were analyzed. There were other symptoms such as speaking, spitting, hugging, and oral and upper-extremity automatisms along with different types of ictal kissing behavior during the seizures. The median duration of all seizures was 106.7 ± 73.73 s. Kissing occurred at a median time of 71 s (1–95 s) after the onset of seizure, and ictal epileptiform discharges usually involved TL during kissing episode.

Conclusion: Ictal kissing is mainly associated with right TL and female gender, although association with left TL may also occur, possibly related to the circuits involving temporo-insular structures.

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1. Introduction

Automatisms are more or less coordinated, repetitive, motor activities that occur when cognition is usually impaired. They often resemble a voluntary movement and may consist of an inappropriate continuation of ongoing preictal motor activity. The patient usually becomes amnesic for these events [1]. Automatisms are categorized as oroalimentary, mimetic, manual or pedal, gestural, hyperkinetic, hypokinetic, dysphasic, dyspraxic, gelastic, dacrystic, vocal, verbal, spontaneous and interactive in the International League Against Epilepsy (ILAE) report of seizure semiology [1]. Some automatisms have lateralizing and localizing value [2] but

their mechanisms and related inner driving sources are still need to be investigated.

Ictal kissing is a rare automatism observed during focal seizures. Only nine patients have been reported previously in the literature. The seizures were localized mainly to TL in all and lateralized to right hemisphere in eight of them [2–7]. Although the clinical significance remains unclear, the occurrence of ictal kissing was found to be associated with common psychological features [7]. We report 16 additional cases from different centers with ictal kissing and reviewed the previously reported nine (4 from our center) cases. In 15 of them, we were able to analyze the semiologic and electroencephalographic features associated with ictal kissing automatism.

2. Methods

Medical records of 20 patients with documented ictal kissing were provided from four epilepsy centers. Demographic, clinical, video EEG, and imaging data, if available, were analyzed and

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reviewed, including the previously reported 9 patients in the literature [2–7].

All patients were resistant to antiepileptic drug medication and subjected to pre-surgical evaluation, including inter-ictal and ictal video EEG, magnetic resonance (MR) imaging, and FDG-PET in 15 of them. Ictal video EEG recordings, which were available from 15 patients, were analyzed considering detailed semiologic and electroencephalographic characteristics such as total seizure duration, timing of kissing automatism, and other features of the seizures.

3. Results

The demographic and clinical data of 25 patients are presented in Table 1. Sixty-four percent of the patients were female. The mean age was 32.9 years (9.9–51 years). The average age at seizure onset was 14.5 years (5–23 years). All patients had focal seizures, with or without secondary generalization. Twenty-two patients were on at least one antiepileptic drug; three patients did not take any drug because they were seizure free. Past history was remarkable with febrile seizures in 7, viral meningitis in 1, neonatal asphyxia in 2, unremarkable or unknown in 16 patients. Neuropsychological evaluation which was available in 13 patients revealed material specific memory deficits in 8 (P1, 2, 4, 10, 11, 12, 13, 19, 20), attention deficits in 4 (P3, 11, 12, 13) and visuospatial deficits in 2 patients (P14, 16) and normal IQ in all. Psychiatric interview disclosed traumatic childhood and unfulfilled affective needs with major depression in 4 patients who were published separately (P1, 2, 3, 4) [7]. Depression was also diagnosed in 1 more patient (P19). However further information related to their psychosocial status cannot be obtained in other patients as they were recruited from different centers.

Nine patients out of 25 underwent surgery, all but one after non-invasive investigations (P1, 10, 12, 14, 15, 17, 21, 24 and 25). One of these surgical patients, who was from Paris, underwent stereoelectroencephalography (SEEG) prior to surgery. Eight patients were seizure free since then. Three patients refused surgery and five were on the waiting list.

Fifty-five ictal video EEGs obtained from 15 patients were analyzed. Ictal kissing was observed in 28 events (50.9%) where TL was involved at the time of kissing in all of them. The seizure onset was located in the TL in 13 patients (10 on the right side, 3 on the left) and was not determined in two patients because of movement artifacts and hypermotor behavior.

Although the majority of the patients had right temporal epileptiform discharges on inter-ictal EEG, they were bilateral in four with a prominence of left TL in two patients (P5, 7). Both inter-ictal and ictal EEG demonstrated left-sided abnormality in two patients (P4, 15). SEEG investigation was recorded in only one patient (P10), who revealed the involvement of temporal pole and lateral temporal neocortex with propagation to the frontal lobe. Unfortunately, kissing automatism was not observed during this intracranial recording. Subdural recording was performed in P25, who was one of the previously reported cases. It demonstrated epileptiform discharges on the right frontal lobe during kissing. The hand dominance was right in 16 right temporal lobe epilepsy (TLE) patients, left in 2 right TLE (P2, 10), left hemispheric dominance in one (P10) of those was demonstrated by the Wada test, and left in only one left TLE patient (P4). The hand dominance was not determined in one patient.

3.1. Semiologic features of seizures

The median duration of all seizures was 76.5 s (5–283 s). Kissing occurred after a median time of 71 s (1–195 s) of seizure onset and was usually preceded by other behaviors such as ictal speech and oral and upper-extremity automatisms. Four patients presented

ictal kissing within the first 35 s (5–33 s) after the onset while it occurred after 35 s in the rest of the patients. Consciousness was partially preserved in 16 seizures arising from right but not left TL.

Aura was described in 13 of 20 patients. Epigastric sensation was the most frequent aura, followed by dreamy state, experiential visual sensations, tinnitus, goose bumps, nausea, smelling unpleasant odors, feelings of confusion, fear, strange sensation, headache, staggers, and anxiety.

The kissing behavior differed from one patient to another. It was not stereotypical even in the same patient. Six patients blew kisses to the air during the seizure. Nearly half of the patients kissed the hands or arm, one third of them kissed the cheek of the individuals around, and one young woman engaged compulsively in lip kissing (P4), which was accompanied by other behaviors such as water drinking, speaking, and hand automatisms. This interesting patient had left-sided hippocampal sclerosis (HS) on MRI with left-sided ictal and inter-ictal epileptiform activity on EEG. Moreover patients did not show any mirthful emotion but either sadness or neutral during kissing behavior.

Seizures were always diurnal in all 20 patients except one (P8) having seizures soon after she fell asleep, which were characterized by staring, lip smacking, and blowing kisses with incomprehensible speech. She had normal MRI and right-sided epileptiform activity on EEG.

3.2. Imaging

MRI scan revealed lesions within the TL in 15 including mesial temporal sclerosis (MTS) in 14 (11 right, 2 left, 1 bilateral), arteriovenous malformation (AVM) in 1. Other observed lesions included cortical dysplasia, posterior cerebral artery (PCA) infarct, AVM, and frontal cortical and subcortical gliosis where it was normal in seven (Table 1). MRI was normal, but histopathology was cortical dysplasia type IIa in one (P21).

FDG-PET was available in 15 patients, which showed hypometabolism at TL in 14 (left in 2, right in 12) and was normal in one (Table 1).

In all patients with abnormal imaging, inter-ictal or ictal EEG abnormalities correlated with the lesional areas.

4. Discussion

Careful analysis of ictal semiology is crucial for delineation of epileptogenic zone. In this context, each ictal phenomenon reflects the involvement of cortical regions by the epileptic discharge from onset to propagation and is investigated in terms of lateralizing and localizing importance. Several semiologic features are described and listed by the ILAE terminology task force, such as motor, nonmotor, and autonomic behaviors [1]. Automatisms are classified as motor symptoms and defined as a more or less coordinated, repetitive motor activity usually occurring when cognition is impaired; the subject does not remember them afterward. The automatisms often resemble a voluntary movement and may consist of inappropriate continuation of ongoing preictal motor activity in the same report [1]. There is a variety of automatisms, such as oral, mimetic, manual, pedal, gesture, and other presentations. Kissing was not mentioned in this report [1], although it may partly fit oral automatisms when lip smacking, lip pursing, chewing, licking, tooth grinding, and swallowing were listed in this group. This may be due to its rare occurrence, as was reflected by only nine patients reported in the literature, four of which were those from 11 patients belonging to our center [2–7].

The mechanism of automatisms has three main speculative explanations. They may emerge as a reaction to internal stimuli, as an activation of a specific motor program with cortical stimulation, or as the removal of inhibitor control as a release phenomenon [8].

Table 1
Electroclinical and imaging features of patients with ictal kissing automatism.

Patients	Gender/ age (yrs)	Age at onset (yrs)	Handed	MRI	PET hypo- metabolism	Inter-ictal EEG epileptiform activity	Ictal EEG	Aura	Associated ictal behaviors
P1,FS*	F/28	16	R	R MTS	None	R > LT	R FT, spreading to bil FT	Epigastric sensation	Religious speech, oral automatism, ictal water drinking, kissing technician's hand, blowing kisses
P2,NM*	F/35	17	L	R MTS	R T	R T	R > LT	Feeling dreamlike and in another place	Oroalimentary automatism, raising L hand, kissing cheek
P3,CG*	M/26	16	R	normal	RT	RT	R > LT	Tinnitus in the R ear, goose bumps	Oroalimentary automatism, speech, blowing kisses, coughing
P4,NI*	F/29	5	L	L MTS	LT	LT	LT	Smelling, bad odors	Speech, L-hand automatism, water drinking, R-hand dystonia, kisses from lip
P5,NG	F/20	9	R	Normal	R T	L > RT	Nonlateralizing and localizing ictal onset late built up at LT	None	Hypermotor automatism, L-hand dystonic posture, coughing, kisses from hand, religious speech, crying
P6,GO	F/51	22	R	R MTS	R T	R T	R FT	Palpitation, paraphasia	Oral automatism, bil upper-extremity automatism, kissing from cheek
P7,MR	F/29	18	R	Normal	None	Bil ant T	None	None	Hand automatism, water drinking, hugging and kissing from cheek
P8,MS	F/24	23	R	Normal	None	R ant T	None	None	Oroalimentary automatism, incomprehensible speech, blowing kisses
P9,BA1	M/46	12	R	L T and O gliosis (PCA infarct)	None	L OT	None	Headache, grayout	Hand automatism, blowing kisses
P10,SF	M/41	10	L	R MTS	R T- F_Insular	R T	R TF	Epigastric sensation, nausea	Oro-alimentary automatism,whistle, gargle, sucking, foreign-language ictal speech, dance-like movements, pedaling, kissing from hand, spitting
P11,FEO	M/31	5	R	Normal	R T	R T	R T	None	Oroalimentary automatism, religious speech, utterance of noises, kissing from hand
P12,GÖ	F/32	12	R	R MTS and O Ulegyria	R T	R T	R T	None	Fondling on arm, kissing from cheek and arm
P13,AD	M/25	10	R	R MTS	R T	R T	R T	Confusion feeling	Right-hand and oral automatism, groaning, kissing from hand, hugging
P14,ZD	F/23	17	R	R MTS	R T	R T	R T	Strange sensation	Staring, kissing from hand, R-hand automatism, L-arm dystonia, religious speech
P15,KT	M/20	9	R	L MTS	L T	L T	L T	Epigastric sensation	Staring, kissing from hand, spitting, vomiting, incomprehensible speech
P16,FG	F/22	9	R	P neuronal migration anomaly	R T	R T	R T, spreading to all R hemisphere	Anxiety	Screaming, fear, hypermotor behavior, oral and R-hand automatism, kissing from cheek
P17,SSK	F/41	9	R	R MTS	None	R T	R T	None	Staring, confusion, kissing from cheek, speech, R-hand automatism, L- arm dystonia
P18,MG	M/44	12	Unknown	Normal	Unavailable	R > LT	None	None	Aggression, hypermotor behaviors, blowing kisses, spitting
P19, BA2	F/32	5	R	Bil MTS	Normal	L T	L > RT	Fear	Oral and hand automatism, blowing kisses
P20, IK	M/37	21	R	L T AVM	None	None	None	Nausea, dry mouth, taste	Speech, blowing kisses, bottom lip
P21. Rashid et al. [5]	F/39	33	R	Normal	R T	R T	R T	Nausea	Tingling arms and legs, oral automatism, blowing kisses, kissing from hand
P22. Rashid et al. [5]	F/46	26	R	R MTS	None	SW, R T	R T, spreading to the R > L F	None	Whimpering, oral automatism, fumbling with R hand, L-arm dystonic posturing, hugging, kissing from cheek
P23. Rashid et al. [5]	F/48	18	R	R MTS	None	SW, R > LT	R FT, spreading to bil FT	Warm, rising sensation	staring, repetitive leg movements, fumbling R hand, oral automatism, blowing kisses
P24. Mikati et al. [4]	M/24	10	R	R MTS	None	Unknown	R T, in some seizures evolving to bil FT	None	Altered consciousness, spitting, repetitive hugging and affective kissing, head and eye deviation to the L
P25. Alsemari et al. [6]	F/29	19	R	R F small cortical and subcortical lesions	R T	SW, R T	R FT	None	R-hand automatism, L-arm dystonic posturing, lip smacking, hugging and kissing her relative and/or attendant nurse with emotional gestures

Abbreviations: M—male; F—female; yrs—years; R—right; L—left; MTS—mesiotemporal sclerosis; T—temporal; OT—occipitotemporal; TF—temporofrontal; FT—frontotemporal; AVM—arteriovenous malformation; bil—bilateral.

Swallowing due to excessive salivation may be an example for the first hypothesis. The second mechanism refers to cortical activation of functionally localized behavior such as simple motor movements usually when the patient is awake. This mechanism may partly explain the emergence of automatism, although some case reports describe electrical cortical stimulation of insular operculum, hippocampus, amygdala, and mesial frontal lobe-induced oral automatism [8,9] and stimulation of hypothalamic hamartomas-induced laughing and crying [10]. By definition, more complex, semi-purposeful automatic movements are seen when the cognition is impaired, and such ictal events including kissing automatism has never been reported to be induced by cortical stimulation. The third mechanism proposing the temporary loss of neocortical control leading to release phenomena may be supported by the presence of similar stereotypic behavior in other neurological disorders such as autism, dementia, and schizophrenia [11,12]. Moreover a dysmnesic phenomena, such as a recurrence of a scene from the past with a strong emotional connotation may also be suggested as a fourth mechanism (P10).

Epileptic seizures may cause not only excitation but also inhibition of ongoing neuronal activity, leading to behavioral dysfunction or disinhibition. Central pattern generators (CPGs) located in the brain stem and spinal cord are essential for survival, such as feeding-, locomotion-, and/or reproduction-inducing behavior or emotions, along with innate motor patterns [13]. Kissing behavior may emerge after loss of inhibition on CPGs as an innate behavior pattern during an ictal event; originate from daily life activities; be inducement of an emotional connection, including demonstration of love for a special person; or be a sign of deferential behavior. Thus, the patients may share similar complex emotional backgrounds. From this perspective, ictal kissing may also be the result of reaction to an internal stimulus occurring as a specific pattern of behavior in relation to loss of conscious control or cortical inhibition.

From a psychological perspective, ictal kissing behavior may be the expression of many kinds of feelings, such as love, respect, and kindness. Except for some African tribes who consider kissing a disgusting gesture, kissing in almost all cultures is a common social interaction starting in early childhood [14]. It may be the reflection of love and sexual desires with emotional bond or of the respect for the person whose hand is kissed. Kissing stimulates the secretion of endorphins during breast feeding when an innate infant behavior resembling kissing induces oxytocin, a facilitator to pair bonding and parental care [15]. Erotic kissing may originate from behaviors related to feeding offspring or in birds feeding their partners before mating. This behavior also includes feelings of safety and trust, which are important features of the pair bond between mother and child [14]. Our psychoanalytic investigation of a small group of patients with ictal kissing revealed traumatic childhood histories and intense unfulfilled affective needs with major depression in all. In that report we proposed that the past negative experiences and need for love may manifest itself through the content of seizures as a desire to kiss or to be kissed [7].

Kissing is an emotional behavior. The limbic system which is composed of the amygdala, hippocampus, parts of the thalamus, and other structures (mammillary body, cingulate gyrus, fornix) has a major role in producing emotion. It is a kind of “primitive core” of the brain strongly associated with emotion. In lower animals, the limbic system helps organize basic survival responses, such a feeding, fighting, or reproduction where an obvious link to emotion remains in humans [16].

Emotional behaviors may change according to the gender, which is attributed to anatomo-functional differences in the mesial temporal region [17]. Expression patterns of some behaviors in male or female could be related to the their various unresolved stages of a love relationship and cultural differences [18]. Although

females consisted of the majority of the patients, there was no association between type of kissing pattern and gender in this study. Overall, we observed kissing from the hand or arm (40%), from the cheek (30%), and from the lips and blowing kisses (30%) without a specific gender predilection. Interestingly, the kissing behaviors were not stereotypical even in the same patient, which may be due to distraction by the environment. Therefore, an association between gender and variable kissing patterns in terms of anatomo-functional link remains speculative.

Kissing automatism is known to be rare but it is not possible to estimate the real incidence in this group of patients as all of them were from a selected population of surgical candidates which is the major weakness of this study. Moreover, the wide difference at the time of occurrence of kissing after a seizure onset, duration of seizures, variety of phenomenology even in a single patient, various underlying etiology, epileptogenic zone extending beyond TL even extra temporal localization implies that the kissing phenomenon is something reflecting a wide network activation by seizure discharge propagation and thus TL involvement rather than the being origin of it. Moreover, “tuning” of the emotional system pre-ictally can be different from one moment to another which is modulated by the limbic system and its connections, the pivotal epileptogenic zone responsible for emotion related ictal signs as mentioned above. This may explain the variability of the occurrence within the seizures and among the patients as well.

The epileptogenic zone was demonstrated to be related to right/language nondominant TL in previous reports [3–6] except one [7]. In this series of 25 patients (including the previously reported 5 patients) it was related to the right side in 16, left side in 4, and unclear in 5 patients due to normal or bilateral MRI and EEG findings. Rashid et al. implied that the importance of the preservation of responsiveness and ictal speech were necessary for ictal kissing to interact sufficiently with the environment [5]; however, the epileptogenic zone in two of our patients was on the dominant TL without speech and responsiveness during the seizure.

Lateralization of brain functions was defined for language-related areas and for different functions, including emotion processing. The right hemisphere was postulated to be responsible for emotional processing [19], whereas valence theory proposes that lateralization depends on the type of emotion; in this view, happiness and affiliated emotions would be processed predominantly by the left hemisphere, and sadness by the right hemisphere [20]. A majority of patients with ictal kissing have shown right-hemisphere involvement without showing and happiness, which may accord with both hypothesis. Not handedness but the type of brain structures [21] was shown to be responsible in emotional processing, which may explain the occurrence of ictal kissing in patients with left-handed or left-hemispheric involvement.

All available video EEG recordings with kissing revealed ictal discharges related to TL even in the patient with parietal cortical malformation who had discharges propagating to right TL at early phase. Three previously presented patients were also reported to have right-TL involvement [5], whereas one with a right-frontal lobe lesion who was explored with subdural electrodes demonstrated the spread to frontal from right mesial temporal who underwent resection of both frontal lesion and and temporal lobectomy with amigdalohippocampectomy [6]. One case with ictal kissing and spitting precipitated by specific patterns was reported to have right mesial temporal lesion and low-grade astrocytoma and remained seizure free after surgery [4]. Temporal lobes were bilaterally involved in three patients (P2, 3,19). There may be some relation to Klüver-Bucy syndrome, which is presented by hyperorality and inappropriate sexual behaviors like the previously reported patient with ictal kissing [5] and ictal hyperorality [22]. The fact that the majority of cases showed one-sided involvement depending on this rare syndrome will not be valid for all patients.

Thus, whether unilateral or bilateral, the majority of patients presenting with ictal kissing in this study correlated with mainly TL involvement according to the scalp EEG, which fails to show the involvement of deep structures such as insula, although SEEG investigation in one patient revealed insula to be included in the epileptogenic area. However, involvement of the insular cortex in almost all subgroups of TL epilepsies which was demonstrated in PET studies [23] is well-known fact, which contributes the mechanistic explanation of kissing automatism.

Several studies demonstrated the prominent role of right anterior insula, a phylogenetically archaic allocortical structure, in processing emotion regardless of handedness and its significance as the principal structure of the cranial nerve system responsible for sentience and self-consciousness [24–26], right hemispheric activation dominance during sadness, and contralateral dominance related to happiness. It is not easy to consider kissing as a negative emotion; however, ictal discharges may have both inhibitory and excitatory effects. However, none of the patients were joyful but either sad or neutral. Therefore, kissing may well be the result of activation of right hemisphere with negative emotions.

Responsiveness may be preserved in patients when the nondominant hemisphere is involved during the seizure. However, they continue to have different types of automatisms, and they can respond to the examiner without being able to recall the event at all. One can argue whether the consciousness is impaired during these seizures. Our understanding and assessment of ictal consciousness, focusing on both subjective and behavioral aspects of seizures, need to be considered here. There have been suggestions that both the internal and external milieu—the former related to the phenomenal qualia of experience, the latter related to behavior—must be taken into account for a better understanding of altered states of consciousness in epilepsy. A bidimensional model, in which any manifestation of conscious experience can be plotted according to the level and contents of consciousness, was proposed when the level axis measures the degree of alertness/arousal, whereas the contents axis measures the vividness of specific experiential phenomena reported by the patient. In a recent paper it was argued that certain seizure types might require more rigorous conceptual models for their characterization, a three-dimensional model that includes a further dimension related to the self, in addition to those of level and contents [27]. A complex behavior such as kissing occurring while the awareness and partial content is preserved during a seizure can also be explained by the impact of a self-component of consciousness according to the new proposal. And while the anterior-dorsal insula was regarded as the final stage of a hierarchical processing, starting in the posterior insula with pure sensory information, then integrating emotional and cognitive valuation, ending in the anterior-dorsal insular region with a full representation of a “sentient self,” the sine qua non of self-awareness [28], invasion of insula with seizure discharge may well be responsible of disruption of self component of consciousness as proposed.

5. Conclusion

Kissing automatism is a complex and rare ictal phenomenon, associated with TLE but in itself reflecting the activation and/or inhibition of networks involved with rather basic innate behavioral patterns more than strict TLE pathology. It is correlated mainly with right-TLE with a predilection in females. However, involvement of dominant TL and male gender is rare but also possible. Although the precise mechanism remains obscure, it may occur as a release or dysmnestic phenomenon rather than a cortical stimulation, possibly due to firing of extensive circuits centered at the insular and TL or as the result of internal stimulation that facilitates CPG with loss of conscious control.

Conflict of interest statement

None of the authors has any conflict of interest to disclose.

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