

Somatosensory Aura in Mesial Temporal Lobe Epilepsy: Semiologic Characteristics, MRI Findings and Differential Diagnosis with Parietal Lobe Epilepsy

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ABSTRACT

Introduction: Somatosensory auras (SSAs) are more usually described in patients with parietal lobe epilepsy (PLE), producing more commonly a localized cutaneous tingling sensation, involving hands and fingers followed by tonic or clonic focal seizures. These usually originate in the contralateral hemisphere. Etiology includes dysplasias, tumours, ischemic or postencephalitic gliosis. However, other focal epilepsies, such as frontal and temporal, may also originate SSAs. Although this type of aura is reported as rare in patients with mesial temporal lobe epilepsy (MTLE), this association has not been systematically studied. **Objectives:** The aim of this article was to describe the cases of four patients with refractory MTLE and SSAs, reporting their clinical characteristics and MRI findings. We discuss the localizing and lateralizing value of SSAs, particularly in the context of MTLE. **Methods and Results:** Four patients with refractory MTLE and SSAs followed-up in the outpatient's clinic at the Epilepsy Section, Universidade Federal de São Paulo, were submitted to presurgical evaluation and corticoamygdalohippocampectomy. MRI in all cases showed unilateral mesial temporal sclerosis (MTS). Regarding seizure semiology, tingling sensation involving the upper extremity was the most prevalent symptom. Three of the four patients had SSAs contralateral to the MTS. Following the SSAs all patients most of the time presented other symptoms such as autonomic or psychic auras evolving to psychomotor seizures. After surgical treatment, two of the patients presented infrequent auras, and two were rendered seizure-free. **Conclusion:** Although rare, SSAs can be present in MTLE. The characteristics of autonomic or psychic auras, psychomotor seizures, neuropsychological deficits, and typical neurophysiologic and MRI findings may help differentiate patients with MTLE from those with PLE.

Key words: somatosensory auras, temporal lobe epilepsy, mesial temporal sclerosis, parietal epilepsy.

RESUMO

Auras somatossensoriais em epilepsia mesial temporal: características semiológicas, achados neurorradiológicos e diagnóstico diferencial com epilepsia de lobo parietal

Introdução: Auras somatossensoriais (AS) são mais frequentemente descritas em pacientes com epilepsia de lobo parietal (ELP), produzindo sensação de formigamento cutâneo envolvendo mão e dedos seguida de sinais motores focais tônicos ou clônicos. Estas crises habitualmente originam-se no hemisfério cerebral contralateral. Displasias, tumores, patologias vasculares ou gliose pós encefalite constituem as etiologias mais prováveis. Outras epilepsias focais, como as de lobo frontal e temporal podem também originar (AS). Embora este tipo de aura seja considerada rara em pacientes com epilepsia mesial do lobo temporal (EMLT),

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esta associação ainda não foi sistematicamente estudada. **Objetivos:** A proposta deste artigo foi descrever quatro casos de EMLT refratária ao tratamento clínico, apresentando AS, reportar as características clínicas comuns, achados de neuroimagem e estudar o valor localizatório e lateralizatório das mesmas. **Métodos e Resultados:** Quatro pacientes com EMLT refratária e AS acompanhados no ambulatório de Epilepsia, Universidade Federal de São Paulo, foram submetidos a avaliação pré-cirúrgica. Sensação de formigamento envolvendo a extremidade superior foi o sintoma predominante em todos. Em três (75%) as auras foram contralaterais à esclerose mesial temporal (EMT). Em todos as AS eram, com maior frequência, seguidas por outras auras autonômicas e psíquicas e evoluíam para crises parciais complexas (crises psicomotoras). **Conclusão:** Embora raras, AS podem estar presentes em EMLT. A associação de auras autonômicas e psíquicas, presença de crises parciais complexas (psicomotoras), déficits neuropsicológicos distintos, além de achados neurofisiológicos e de neuroimagem podem diferenciar pacientes com EMLT daqueles com ELP.

Unitermos: auras somatossensoriais, epilepsia mesial do lobo temporal, esclerose mesial temporal, epilepsia de lobo parietal.

INTRODUCTION

Auras are subjective ictal phenomena that may precede an observable seizure. Somatosensory auras (SSAs) are specific somatic sensations described as tingling, numbness, electric shock sensation, pain, sense of movement, or desire to move⁽²⁾. SSAs frequently occur in patients with parietal lobe epilepsy (PLE). The initial description of paraesthetic seizures was done by Jackson in 1863^(apud 9). Following Jackson, Penfield, in the Montreal school, demonstrated that the characteristic spontaneous seizures could be reproduced by electrical stimulation of primary, secondary and supplementary somatosensory areas. In monkeys three somatosensory areas have been described (Figure 1).

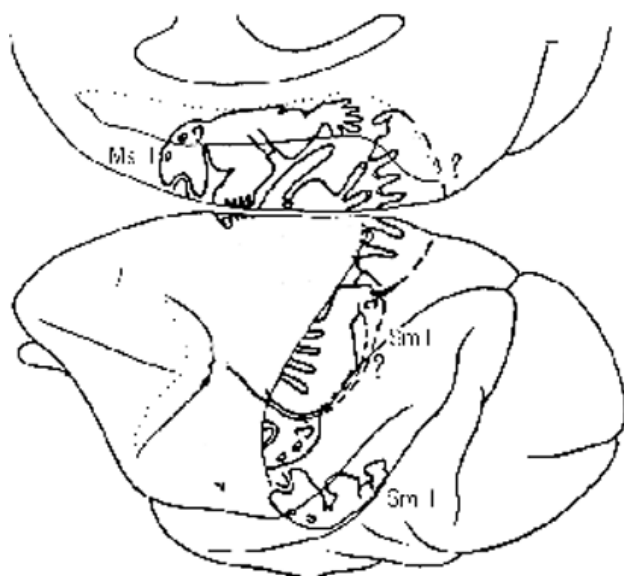


Figure 1. Somatosensory areas in monkeys: Sm I Somatomotor primary area; Sm II Somatomotor secondary area; MSII: Supplementary motor area. In monkeys as in men Sm II is located in the upper bank of Sylvius fissure adjacent to the insula. Modified from Brodal⁽³⁾.

Unilateral paraesthetic seizures, with or without “Jacksonian march”, are considered the clinical expression of involvement of contralateral somatosensory primary area⁽⁹⁾. Bilateral paraesthetic seizures, with or without acoustic sensation, are due to involvement of secondary or supplementary sensory areas^(9,15). The most frequent somatic topography in unilateral SSAs is the hands and fingers⁽⁹⁾. Arm, foot, leg, face, head and trunk may also be the loci of initial symptoms. These can spread during the seizures, with progressive march in the cortical primary sensory area (SI), or remain located^(9,11,13). Jacksonian march usually involves the upper extremity. Rarely, somatic sensation is diffuse or generalized. Tingling is the most frequent sensation in the SSAs due to focal epilepsy, constituting about half of the cases⁽¹³⁾. Patients with somatosensory symptoms usually present focal motor seizures, with clonic, tonic or postural movements. Negative motor, hypermotor and versive seizures are less common⁽¹³⁾. Psychomotor seizures, typical of temporal lobe involvement, may be observed in up to 30% of the patients with SSAs.

Tumors, dysplasia, vascular pathology, traumatic brain injury and postencephalitic lesions have been found in patients with focal epilepsy and SSAs. Although paraesthetic seizures have been reported in parietal epilepsy, they can be present in other focal epilepsies. Frontal and temporal lobe epilepsies may also present SSAs^(7,13).

Temporal lobe epilepsy (TLE) is the most common focal epilepsy in adults. Mesial temporal lobe epilepsy (MTLE) corresponds to about 60% of the cases, and is a well-defined syndrome characterized by autonomic, epigastric or psychic auras, followed by complex partial seizures, with behavioral arrest and staring, oral or hands automatisms, frequently associated with dystonic posturing of the upper extremity. Lateralizing ictal and postictal symptoms in patients with MTLE, associated with neurophysiologic and imaging data may enhance diagnostic

accuracy. Nevertheless, semiologic characteristics remain the essential criteria. More recently, using stereotactically implanted depth electrodes in the posterior part of the insular cortex in humans, Ostrowsky et al. (12) demonstrated the representation of somatic sensation in this area of the brain. SSAs are reported to be rare in TLE, occurring in only 1.7% to 4.5% of cases⁽⁴⁾. In this study, we describe four patients with MTLE who reported SSAs (Figure 2).



Figure 2. Insula. Anterior, medium and posterior short gyri and anterior and posterior long gyri as well as the Heschl gyrus. Stimulation of the posterior insula produces somatosensory symptoms⁽⁹⁾.

PATIENTS AND METHODS

Four patients were investigated. All of them had complete semiologic data, with detailed clinical history, seizure description by themselves and their families, neurological, psychiatric and neuropsychological examinations and high resolution MRI study. Prolonged interictal and ictal video-EEG recordings of at least three seizures were carried out in all. In every case an epileptologist or a specialized nurse

examined the patient during and immediately after seizures. The age at seizure onset and duration of epilepsy were also noted. We included patients with SSAs, excluding those with purely visceral auras or cephalic sensations. These events were recorded during video-EEG, and correlated with image results and specialists evaluations.

RESULTS

Three of the four patients were females. The onset of epilepsy ranged from 3 to 17, and mean epilepsy duration was 25.5 years (Table 1). The neurological examination was normal in two patients, and in two showed memory deficits. None of them presented sensorimotor deficits.

SSAs types described by the patients were tingling (in two), numbness and thermal sensation such as being cold. The initial somatotopic localization was hand (in two) and leg and trunk. In one the aura was confined (patient 4 – tingling in left leg), and in three there was spreading of somesthetic symptoms. Patient 1 had numbness in left trunk followed by involvement of the whole left side. Patient 2 had cold sensation in the left hand followed by “Jacksonian march” (upper limb – face-lower limb). Patient 3 referred tingling in the right hand, subsequently involving the unilateral upper limb and contralateral hand. In three cases auras were contralateral to the ictal onset zone, and in one, ipsilateral. In three (patients 2, 3 and 4) psychomotor seizures were the subsequent manifestation, in all with dystonic posturing contralateral to the ictal onset zone. Seizures in patient 1 initiated with a scream followed by gross motor automatisms involving both upper and lower limbs and afterwards behavioral arrest. All the patients exhibited other auras. Patient 1 and 2 related ictal fear; patients 2 and 3 had autonomic aura (epigastric sensation) and patient 3 referred “jamais vu” (for more details see Table 2).

Table 1. Clinical and epidemiologic data and postoperative outcome

Patient	Age	Gender	Duration	Ictal*	Aura	MRI*	Surgery	Outcome
1	34	F	31	RTL	Numbness in trunk, half left body	R MTS	01/2006	Seizure free for 7 months
2	39	F	22	LTL	Sensation of cold in left hand, left mouth, left leg	L MTS	02/2006	Seizure free for 6 months
3	40	M	25	LTL	Tingling in right hand, ascending in right arm or involving both hands	L MTS	11/2002	Seizure free for 3.7 y, still presenting auras
4	37	F	24	RTL	Tingling in left leg	R MTS and right parietal calcification	02/2004	Seizure free for 2.5 y, still presenting auras

* R: right; L: left; MTS: mesial temporal sclerosis; RTL: right temporal lobe; LTL: left temporal lobe.

Table 2. Topographic distribution and clinical manifestations of somatosensory auras

Patient	Focal aura	Maximal spread	Sensation	Other auras	Ictal zone onset	Subsequent seizure manifestation
1	trunk	whole side	numbness	+	contralateral	hypermotor and psychomotor
2	hand	Jacksonian march	cold	+	ipsilateral	psychomotor
3	hand	upper unilateral – upper contralateral	tingling	+	contralateral	psychomotor
4	leg	–	tingling	+	contralateral	psychomotor

High resolution MRI showed signs of mesial temporal sclerosis (MTS) in all patients. MRI in patient 4 also showed a calcification in the right parietal region. Neuropsychological evaluation showed that only patient 1 had a mild mental deficiency measured through subtests of WAIS-III (IQ = 69).

Localization of the seizure onset was determined by clinical semiology along with interictal and ictal

electrophysiological studies and MRI (Figures 3 and 4). The epileptogenic temporal lobe was the right in patient 1 and 4 and the left in 2 and 3.

Psychiatric evaluations were available for all before and after surgery. Patient 1 had a history of 5 suicidal attempts, with no axis I diagnosis according to DSM IV-TR. She was given up for adoption at birth and suffered a violent home environment, running away at age 13 and getting married to her present partner. As for her complicated past history she has had psychological follow-up after surgery. The remaining patients did not present psychiatric alterations.

All patients underwent corticoamygdalohippocampectomy. In this surgical technique 2-3 cm of the left and 4-5 cm of the right neocortex are resected; most of the amygdala is aspirated, leaving 20% of amygdalian tissue close to the internal capsule, while 2-3 cm of hippocampus is also removed. As for the surgery outcomes all four patients had good results. Patients 1 and 2, in a short follow-up (< 1 year), did not report any auras or seizures whatsoever. Patient 3 now reports focal somatosensory symptoms restricted to the right hand and patient 4 seldom refers auras.

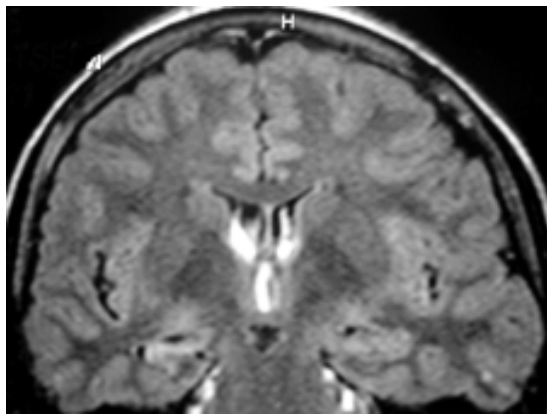


Figure 3. Coronal FLAIR MRI showing right hippocampal atrophy (Patient 1).

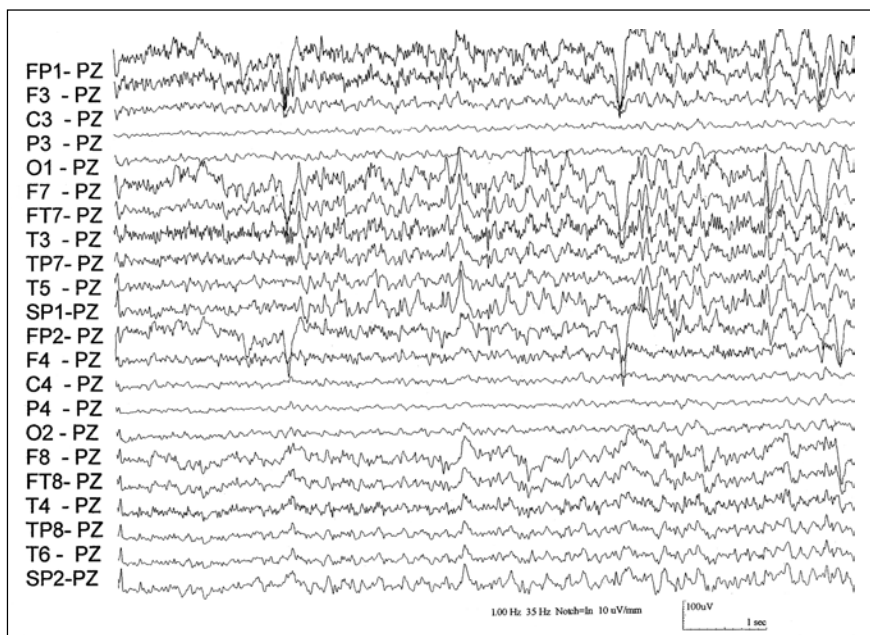


Figure 4. Ictal rhythm developing over the left temporal region and left parasagittal area, accompanying aura described by cold sensation on left hand (Patient 2). This was the only aura ipsilateral to the mesial temporal sclerosis.

DISCUSSION

Although rare, SSAs associated with MTLE have previously been reported in the literature. Six cases of MTLE with SSAs were described by Tuxhorn⁽¹³⁾ and four by Erickson et al.⁽⁴⁾. Psychomotor seizures were the most common seizure type after SSAs in these cases. The data of our study are similar. However, in our series in one case the initial seizure was characterized by hypermotor automatisms preceding the psychomotor seizure. Kim et al.⁽⁷⁾ revised PLE semiology and reported that in this form of epilepsy focal tonic or clonic motor seizures are the most frequent seizure type, followed by automotor and dialeptic seizures. Generalized tonic-clonic seizures are more frequent in PLE than in TLE.

The possible association of other auras (autonomic, psychic, olfactory and gustatory) with SSAs in TLE patients is described in the current literature. Patients with PLE and SSAs may present various types of aura (more frequently vertigo and visual)⁽⁷⁾. In our series, all patients presented SSAs associated with classic temporal lobe auras such as epigastric, fear, cephalic and other psychic auras.

Regarding ictal onset zone, three cases of our sample presented contralateral auras (two in limbs, one axial) and one, ipsilateral (limb). This finding was concordant with various cases previously presented. Erickson et al.⁽⁴⁾ noted that ipsilateral aura could be correlated with head topography, differently from our work and from the literature^(8,13,15). SSAs occur in one-third to half PLE cases. Contralateral limb is affected in 80% of them^(7,13).

Neither of the patients cited painful aura. Nair et al.⁽¹¹⁾, reported painful aura in TLE, ipsilateral or poor somatotopy, involving the secondary somatosensory cortex. Of twenty-five patients, fourteen had EEG foci in temporal lobe. Only three cases presented auras classically associated to temporal lobe as abdominal, funny feeling and fear.

Several authors suggest the participation of other structures in the genesis of SSAs. Wunderlich et al.⁽¹⁵⁾, in study of bilateral SSAs and TLE, found the main area of hypometabolism in TLE was located adjacent to the acoustic and secondary somatosensory cortex. This may explain the non-localized initial nature of the aura. However, most patients of this study presented auras located in limbs. Ostrowsky, of Mauguière group in France, investigated forty-three cases of TLE by stereotactic electrodes implanted in insula⁽¹²⁾. They reproduced painful and non-painful somesthetic sensations by direct stimulation of insula, thus demonstrating the role of this structure in these auras. Non-painful SSAs in limbs were evoked by stimulation in posterior insula, and in midline structures (lips, tongue) by stimulation of more anterior

sites. The somatotopic representation in insula of non-painful auras described above could suggest topographical diagnosis in our patients. The fact of reproducing thermal sensations and tingling through the stimulation of the posterior region of insula however, does not prevent that intimate connection of this area with the secondary somatosensory area, located in its neighborhood, could be the auras real origin.

Video-EEG and MRI findings differ in PLE patients when compared to those with TLE. In PLE, ictal EEG localization is often poor and false localization in temporal region is frequent. These patients frequently exhibit inconsistent clinical picture^(7,9,13). MRI showing extratemporal lesions may worsen the prognosis⁽⁷⁾. Cortical dysplasia is the most frequent pathological finding. In MTLE, scalp EEG reveals paroxysmal abnormalities during prolonged monitoring in 96% of patients, and in 94% of them sharp waves are maximum over the anterior temporal region. Ictal EEG changes are rarely detected at the time of clinical seizure onset, but lateralized buildup of rhythmic seizure activity during the ictal period occurs in 80% of patients⁽¹⁴⁾. MTS is the most common underlying pathology for refractory temporal lobe epilepsy. MRI exhibits hypointensity signal in T₁-weighted and disruption of the internal structure of the hippocampus, T₂-weighted increased signal and hippocampus atrophy⁽⁶⁾.

The four patients of this series had seizures registered by prolonged video-EEG monitoring with ictal onset over the temporal region and MRI showing signs of unilateral MTS.

As to surgical outcome, two were rendered seizure-free in a follow-up of six months and two still present rare auras (Engel class IB). In patient 3 there was a modification of the semiologic pattern, the aura becoming restricted to the right superior member. In patient 4 the semiology did not suffer modification, although they have been less frequent than before surgery.

The importance of this case reports is that auras with somatosensory manifestations do not necessarily indicate extratemporal seizure onset and SSAs may be associated with temporal lobe epilepsy. It may be bilateral or unilateral and should not serve as a deterrent to temporal lobe resection in patients with refractory epilepsy in whom semiologic, electrophysiologic, and imaging findings clearly support a unilateral temporal lobe seizure focus.

In conclusion, SSAs may be reported by patients with MTLE, although they are a rare event in this specific population. The differential diagnosis should be done with PLE. The failure to recognize this association may lead to delay in adequate diagnosis and treatment and worsen prognosis. Further studies are necessary in order to elucidate the role of the insula and its connections in the generation of SSAs in MTLE patients.

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