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INSULAR EPILEPSY

SIMILARITIES TO TEMPORAL LOBE EPILEPSY

CASE REPORT

ARTHUR CUKIERT, CASSIO FORSTER, MARIO S.D. ANDRIOLI, LEILA FRAYMAN

ABSTRACT - Insular epilepsy has been rarely reported and its clinical and electrographic features are poorly understood. The electrographic study of the insula is difficult since it is hidden from the brain surface by the frontal and temporal lobe. A 48 years-old woman started having simple partial autonomic and complex partial seizures with automatisms and ictal left arm paresis 8 years prior to admission. Seizure's frequency was 1 per week. Pre-operative EEG showed a right temporal lobe focus. Neuropsychological testing disclosed right fronto-temporal dysfunction. MRI showed a right anterior insular cavernous angioma. Intraoperative ECoG obtained after spliting of the sylvian fissure showed independent spiking from the insula and temporal lobe and insular spikes that spread to the temporal lobe. The cavernous angioma and the surrounding gliotic tissue were removed and the temporal lobe was left in place. Post-resection ECoG still disclosed independent temporal and insular spiking with a lower frequency. The patient has been seizure-free since surgery. Insular epilepsy may share many clinical and electroencephalographic features with temporal lobe epilepsy.

KEY WORDS: epilepsy, insula, electrocorticography, cavernous angioma.

Epilepsia insular: similaridades à epilepsia do lobo temporal - relato de caso

RESUMO - A epilepsia insular tem sido raramente relatada e suas características clínicas e eletrencefalográficas são pobremente conhecidas. O estudo eletrográfico da ínsula é difícil já que ela se encontra recoberta pelos lobos frontal e temporal. Uma paciente, de 48 anos, começou a ter crises parciais simples autonômicas e crises parciais complexas com automatismos e paresia crítica de membro superior esquerdo 8 anos antes desta internação. A frequência de crises era de 1/semana . O EEG pré-operatório mostrou foco temporal direito. Testagem neuropsicológica demonstrou disfunção fronto-temporal direita. RMN demonstrou cavernoma insular anterior direito. A eletrocorticografia intraoperatória obtida após a abertura da fissura sylviana demonstrou a presença de espículas independentes na ínsula e no lobo temporal, além de descargas que se originavam na ínsula e espraiavam ao lobo temporal. O angioma cavernoso e a área gliótica ao seu redor foram removidos e o lobo temporal foi deixado em seu lugar. A eletrocorticografia após a ressecção ainda demonstrou a presença de descargas nestas regiões, em menor frequência. A paciente está sem crises desde a cirurgia. A epilepsia insular pode compartilhar diversos aspectos clínicos e eletrográficos com a epilepsia do lobo temporal.

PALAVRAS-CHAVE: epilepsia, insula, eletrocorticografia, angioma cavernoso.

Insular epilepsy has been rarely described in the literature. The insula is one of the five cerebral lobes and its cortex is situated deeply within each hemisphere. It is overlayed by the frontal and temporal neocortex and this explains how difficult it should be to get reliable EEG sampling

Serviço de Cirurgia de Epilepsia, Hospital Brigadeiro, São Paulo SP, Clínica de Diagnóstico e Terapêutica das Epilepsias-SP, São Paulo SP. Aceite: 9-outubro-1997.

Dr. Arthur Cukiert - R. Nova York 744/131 - 04560-001 São Paulo SP - Brasil.

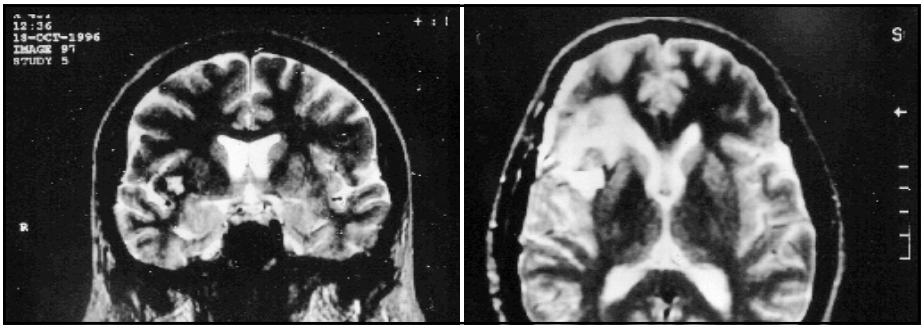

Fig 1. Left: Pre-operative coronal T2-slice showing a right insular cavernoma. Right: Post-operative T2-slice showing the complete removal of the lesion and the surrounding gliotic/hemosiderotic tissue. There was no lesion to the temporal lobe.

from insular cortex and to define an "insular epileptic syndrome" as has been done with temporal lobe epilepsy[6]. Adequate sampling from the insula can only be obtained by depth or subdural electrodes' implantation or acute intraoperative electrocorticography. Depth or subdural electrodes implantation of the insula faces some technical problems. Since depth electrodes should travel through the sylvian fissure, they should be implanted with especial care for the middle cerebral artery branches within the sylvian fissure and requires 3-D angiograhy and MRI or CT integration to be carried out. Chronic implantation of the insula with subdural eletrodes requires the split of the sylvian fissure and placement of strip electrodes within this narrow space, which is on the other hand extremely sensitive to the eventual mass effect of the electrodes themselves. Both procedures have been rarely carried out[3]. The easiest way of sampling the insula is by means of intraoperative electrocorticography (ECoG) after the split of the sylvian fissure. There is a time and spatial limitation (exposed cotex) while performing ECoG; yet, reliable electrophysiological data can be obtained[7].

This paper describes a case of insular epilepsy as defined by clinical, imaging and invasive electrophysiological data and discusses the similarities of insular and temporal lobe epilepsy.

CASE REPORT

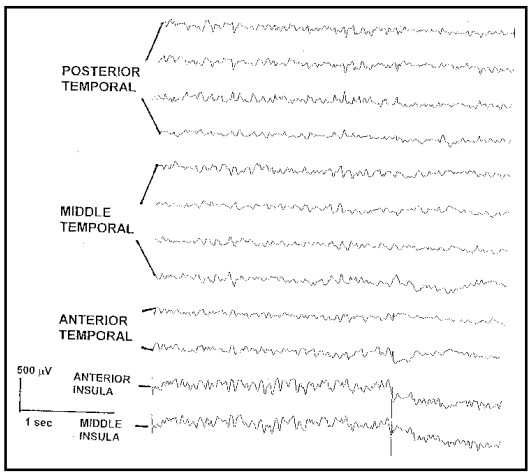

Fig 2. Pre-resection digital ECoG showing insular spiking with limited spread to the anterior temporal lobe.

MARV, a 48 years-old female started having simple partial autonomic and complex partial seizures by the age of 40. Autonomic seizures were characterized by an ascending epigastric sensation and was followed by complex partial seizures with right arm automatisms and left arm paresis. Seizure's frequency was of 3-4 per month. There were 2 tonic-clonic generalized seizures over the last 3 months but these were sporadic. Pre-operative EEG investigation including zygomatic electrodes showed a clear right anterior temporal lobe focus. Neuropsychological testing disclosed a non-dominant fronto-temporal dysfunction. MRI showed an anterior insular cavernoma (Fig 1, left). Cerebral angiography was normal.

This patient was submitted to a right fronto-temporal craniotomy under general anesthesia. After the opening of the dura, the

sylvian fissure was widely split and electrocorticography electrodes were placed over the exposed surface of the insula and over the frontal and temporal lobes. This referential recording disclosed the presence of independent insular and temporal lobe spiking and of discharges clearly originating from the insula and spreading into the temporal lobe (Fig 2). The insular cavernoma and its surrounding gliotic and hemosiderotic tissue were removed using microsurgical techniques. The temporal lobe was left in place and there was no damage to any of its surfaces. A post-resection ECoG recording disclosed a pattern similar to the pre-resection one but with an obvious reduction of the spikes' frequency. There was no post-operative complications. A post-operative MRI documented the complete removal of the cavernoma and the absence of surgically induced temporal lobe lesions (Fig 1, right). This patient is seizure free 6 months after surgery.

DISCUSSION

The pre- and post-operative data presented here strongly suggest that this patient had insular epilepsy. Many of the clinical features would suggest temporal lobe epilepsy and actually, may be the result of the spread of the ictal insular activity directly into the temporal lobe or through arcuate and uncinate bundles' fibers[4]. The simple partial autonomic seizures were similar to those described by temporal lobe epilepsy patients. This type of autonomic seizures has been described by the stimulation of both the amigdala (within the temporal lobe)[1,6] and the insula[2,3,9], but has been said to be more frequently obtained from the mesial temporal lobe structures. This is also true for the ictal complex partial seizures phenomena of ipsilateral automatisms and contralateral upper limb paresis which have been documented to be present in many patients with temporal lobe seizures arising from the hippocampus. On the other hand, even in temporal lobe epilepsy, the distonic/paretic posturing is said to be related to the involvement of ipsilateral basal ganglia relays and not to the activation of the temporal lobe itself.

Involvement of the insular cortex in temporal lobe epilepsy has also been considered in the early days of temporal lobe epilepsy surgery[5]. It has been shown that insular cortex removal in patients with temporal lobe epilepsy did not increase the surgical success rate in relation to seizures and that this procedure carried out an unacceptable morbidity and mortality rates[8]. On the other hand, the involvement of the temporal lobe in insular epilepsy has been poorly studied and virtually nothing is known about it. The presence of independent temporal lobe spiking during ECoG would suggest that this lobe was involved in the epileptogenic proccess in this patient with insular epilepsy.

Many cavernomas, especially the smaller ones, are not seen in regular CT scans and are only detected by MRI scanning. This may also be true for some low-grade gliomas. The clinical and surface EEG data from this patient (disregarding MRI data) would suggest a temporal lobe epilepsy. This raises the intriguing question of how many insular epilepsies were misdiagnosed as temporal lobe epilepsy in the past and how this eventual misdiagnosis influenced the surgical results in the pre-MRI period.

Insular epilepsy shares many clinical and EEG features with temporal lobe epilepsy. Brain imaging with MRI is probably the main available tool to distinguish between them.

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