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Case Report

Sutton's law in epilepsy: Because that is where the lesion is

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ABSTRACT

Successful epilepsy surgery requires unambiguous identification of the epileptogenic zone. This determination may be a challenge when the pre-surgical evaluation yields conflicting data. We evaluated an adult patient with a right insular mass, but a seizure semiology, interictal EEG, and ictal EEG, suggesting left temporal lobe epilepsy. Resection of the mass, a ganglioglioma, resulted in seizure freedom and disappearance of interictal left temporal lobe epileptiform discharges. This case illustrates the principle that in localization-related epilepsy, the money is usually in the mass.

1. Introduction

People rob banks because they are certain to contain money. In patients with localization-related epilepsy, a focal lesion, and a pre-surgical evaluation inconsistent as to seizure onset zone, the "money" is most likely to be in the lesion. We present a patient with a right insular mass, but a seizure semiology, interictal EEG, and ictal EEG, suggesting left temporal lobe epilepsy.

2. Case report

A 28-year-old right-handed man with a history of headache had an approximately two-year history of spells that had evolved in semiology and severity. They began as paroxysmal numbness and "tightening" in the left lower leg that lasted seconds and occurred up to eight times per day. Over the next two years, this sensation increased in intensity and, during longer episodes lasting tens of seconds, spread proximally in Jacksonian fashion to include the entire left hemibody. Three months prior to evaluation at our center, he developed stereotypical episodes lasting about 1 min with a semiology suggestive of temporal lobe complex partial seizure. They were characterized by retained consciousness, unresponsiveness, dysphasia progressing to complete aphasia, and manual automatisms, with post-ictal amnesia and confusion lasting up to tens of minutes. He was able to recall that at least some of these episodes began with the sensation of ascending numbness and tightness. Initially occurring up to three times per day, they decreased to several times per week with the introduction of topiramate titrated to 400 mg/day.

Four habitual seizures with a semiology as described above were recorded in a 10-hour interval during video-EEG monitoring. If the patient was talking at seizure onset, his speech progressed from coherent to intelligible but non-sensical words, to non-word sounds, and to muteness over about 15 s. If he was not talking at seizure onset, the only sounds were occasional mumbling.

Interictal EEG was notable for left anterior temporal spike-wave discharges and polymorphic delta slowing, increased during sleep. Ictal EEG began with low voltage (<10 μV) rhythmic 5.5- to 7.5-Hz theta activity over the right frontotemporal region, maximal at electrodes F4/F8, evolving over several seconds in amplitude, frequency and distribution to the right frontocentral region, maximal at electrodes F4/C4 (Fig. 1). Within 5 to 7 s of seizure onset, the right hemisphere ictal pattern was replaced by a left temporal ictal pattern consisting of higher amplitude (50 μV), sharply contoured rhythmic 4-Hz theta activity maximal at electrodes F7/T3 but with a broad field, which evolved in frequency and amplitude but remained maximal in the left temporal region. Post-ictally, there was diffuse voltage attenuation lasting several minutes, followed by several more minutes of increased left anterior temporal polymorphic delta slowing.

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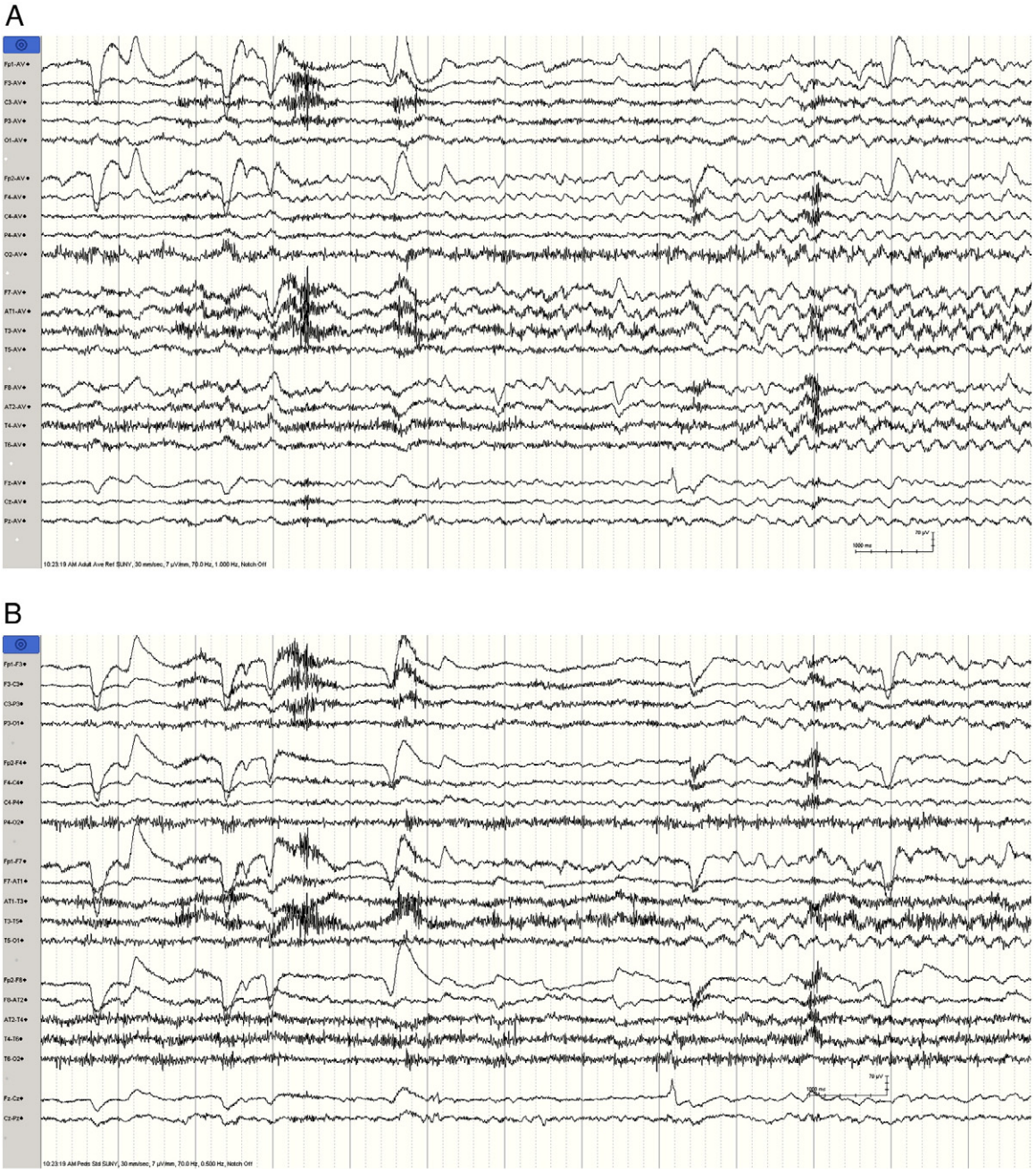

Fig. 1. EEG at onset of a typical seizure. (A) Average reference montage illustrating rhythmic theta activity at F4 beginning in the 3rd second which spreads to C4 and then to the left temporal lobe. (B) AP bipolar montage where the initial rhythmic theta activity is best seen at electrode derivations F4–C4 and C4–P4 before propagating to the left temporal electrode chain.

Brain MRI with and without gadolinium contrast revealed a small cystic lesion with mild enhancement in the region of the right insula (Fig. 2). No other abnormalities were present, and in particular, the temporal lobes were normal. The tumor was completely resected without complication. Histopathology was consistent with ganglioglioma (Fig. 3). By the ninth post-operative month, antiepileptic medications (levetiracetam and lamotrigine) had been discontinued without seizure recurrence. Forty-eight-hour ambulatory EEG obtained 11 months after surgery revealed a right frontotemporal breach rhythm and minimal right mid-temporal theta and delta slowing. At last follow-up, 16 months after tumor resection, he remained seizure free. Medications included valproic acid for headache prophylaxis and paroxetine.

3. Discussion

Gangliogliomas are neoplasms composed of neuronal and glial elements. The most frequent site is the temporal lobe [1]. Although gangliogliomas account for a small proportion of brain neoplasms, they are the most common cause of tumor-related refractory epilepsy [2], particularly in young patients such as the one described here [3].

Guillaume and Mazars [4] and Penfield and Jasper [5] described the typical semiology of insular seizures beginning with somatosensory symptoms possibly combined with motor and visceral components. Isnard and colleagues [6] analyzed ictal symptoms in six patients with insular seizures and suggested five characteristic