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Case report

Epilepsy surgery outcome in temporal lobe cavernoma and multiple sclerosis

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1. Introduction

Seizures are the most common presenting symptom in patients with supratentorial cerebral cavernomas (CC) and can progress to medically refractory epilepsy in approximately 40% of cases [1]. While most studies of CC examine the risk of intracranial hemorrhage, eliminating the seizures can significantly reduce disability and improve quality of life. However, this important benefit often remains an underappreciated treatment goal in managing CC [2]. The literature documents several studies of patients with well-established refractory epilepsy secondary to CC, where surgery of the cerebral lesions significantly improved seizure control [1,3].

We report a patient with pharmacoresistant epilepsy due to cavernoma in the right temporal lobe and comorbid multiple sclerosis (MS) who was considered as a candidate for surgical treatment. The patient underwent anterior partial resection of the right temporal lobe. After the surgery, he remained seizure free but with severe amnestic deficit and devastating MS progression.

2. Case report

The patient is a 45-year-old right-handed man with 10 years of epilepsy history, and unremarkable perinatal and family history. He began experiencing seizures at 35 years of age, at the rate of one seizure per month, characterized by a sudden and strange cephalic feeling ("a storm is coming") followed by loss of consciousness. His spouse, who witnessed the seizures, described a sudden onset of motionless staring, followed by fumbling and fidgeting movements of both arms, and lip smacking, chewing, and swallowing. These seizures infrequently continued to include loss of postural control and bilateral limb jerking. The patient regained reactivity 1 min after habitual seizure but remained confused for the next few minutes.

The patient was initially treated with a titrating dose of carbamazepine (1200 mg/day) without seizure reduction. A dose of 750 mg/day of valproic acid was added and the seizures came under control. Standard scalp EEG showed interictal sharp waves in the right temporal region. Brain magnetic resonance imaging (MRI) disclosed a cavernoma in the mesial part of the right temporal lobe. The seizures recurred after 2 years (42 years of age) at a significant frequency of 1–2 per week. Further titration of carbamazepine (1600 mg/day), valproic acid (2000 mg/day), and the addition of levetiracetam (3000 mg/day) failed to improve seizure control.

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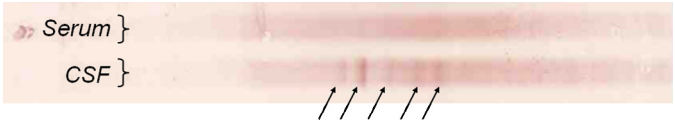

Fig. 1. Parallel cerebrospinal fluid (CSF) and serum examination showed intrathecal presence of oligoclonal bands (*black arrows*).

One year later, numbness and weakness of the right leg and arm occurred. Consecutive brain MRI showed several T2 hyperintensity lesions in the white matter in addition to a cavernoma in the right temporal lobe. A diagnosis of multiple sclerosis (MS) was established after the analysis of cerebrospinal fluid (CSF) showed oligoclonal bands (Fig. 1). Administration of high doses of intravenous corticosteroids over 5 days promoted full recovery and the patient remained in clinical remission for the next 2 years. However, no influence on seizure frequency or seizure semiology was observed.

The patient was referred to our telemetry unit and underwent long-term video-EEG monitoring with scalp electrodes. Three typical seizures were recorded over 5 consecutive days: after the patient felt the usual aura, he become motionless for a few seconds and then exhibited oroalimentary and exploratory automatisms. In all 3 seizures, ictal EEG showed seizure onset over the right anterior temporal region in the form of rhythmic 5 Hz theta activity. Presurgical MRI re-examination confirmed the left temporal cavernoma as well as the bilateral white matter demyelinating lesions (Fig. 2).

The results of the patient's neuropsychological assessment showed a verbal IQ score of 110 and a nonverbal IQ score of 111 with mild prefrontal dysfunction. The Edinburgh Handedness Inventory showed that he was strict right-handed. As the patient had no left-handed family members, we concluded that epileptogenic zone was in non-dominant temporal lobe. Hence, we did not consider a functional MRI language testing.

The patient's case and the possibility for surgical treatment were discussed at a multidisciplinary meeting. There were some questions regarding the possible influence of MS on postoperative outcome and the influence of surgical treatment on the course of MS, respectively. The conclusion was that epilepsy due to the right temporal mediobasal cavernoma significantly decreased the patient's quality of life, and the patient was referred for epilepsy surgery.

The patient underwent epilepsy surgery in April 2011. Anterior partial resection of the right temporal lobe was performed, including amygdalo-hippocampectomy and lesionectomy. The histopathological investigation proved, in correlation with MRI finding, the presence of a cavernoma. In the immediate postoperative course, severe amnestic syndrome with global declarative memory deficits were noticed.

The patient was unable to retain any new verbal information (names of new hospital staff or the information that he read each day in the newspapers). He also demonstrated impaired retrograde episodic memory but intact memory for information regarding his semantic knowledge and previously learned skills. His short-term verbal memory and repetitive speech were normal.

A brain MRI performed 10 days after the surgery revealed postoperative tissue defect in the right temporal region and the existence of new demyelinating lesions in the left thalamus (Fig. 3).

One year after the surgery, the patient was seizure free and classified as Engel I. In the meantime, several relapses of MS occurred with progressive neurological deterioration and severe functional deficit. As high doses of intravenous corticosteroids failed to improve functional disability, mitoxantrone was introduced. The control neuropsychological assessment showed that verbal memory function was significantly decreased in comparison to presurgical scores, with poor recall and poor recognition of previously presented information. The patient's inability to form new episodic memories remained the most significant behavioral symptom.

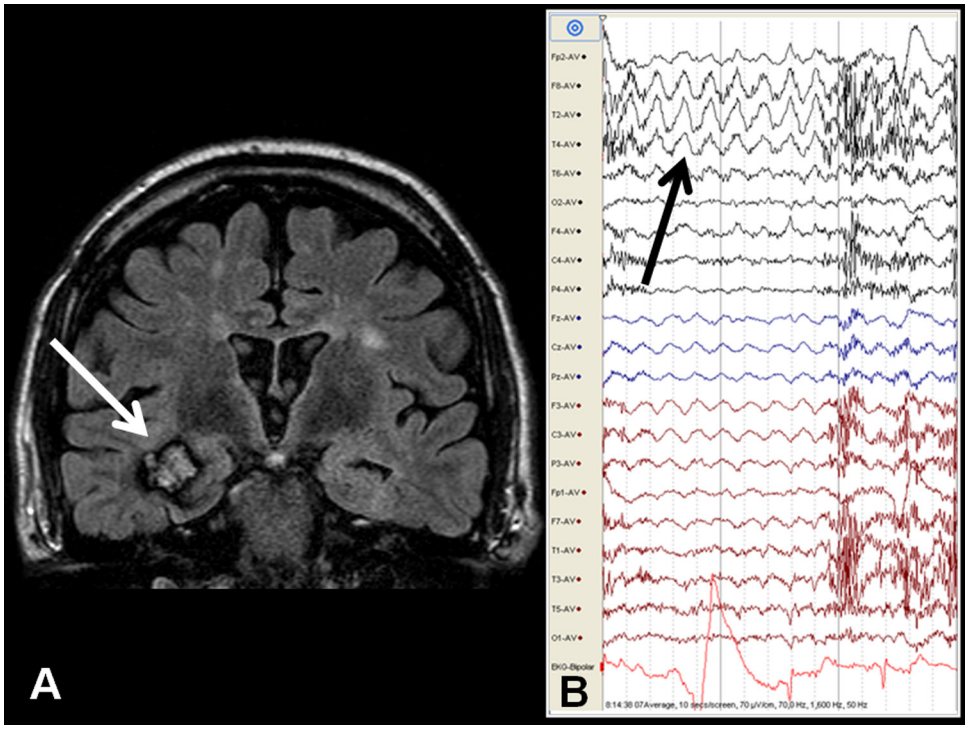

Fig. 2. Presurgical coronal FLAIR image (1.5 T) showing a cerebral cavernoma in the mesial part of the right temporal lobe (*white arrow*) and bilateral demyelinating lesions in white matter (A). Ictal EEG revealed seizure onset zone (*black arrow*) over the right anterior temporal region (B).