Case Report
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Reversible Splenial Corpus Callosum Lesion and Carbamazepine

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Abstract

Background: Reversible splenial lesion syndrome (RESLES) at corpus callosum (CC) has been defined in many clinical conditions. Although the etiopathogenesis of transient focal lesions in the CC splenium has not been clarified, many theories remain on the agenda, especially in epilepsy patients.

Case presentation: A 38-year-old male patient with idiopathic generalized epilepsy was admitted to the emergency department with complaints of frequent seizures. It was reported by his relatives that he had stopped carbamazepine treatment for the last three days. After his last epileptic attack, he had been headache, nausea, vomiting, decreased visual acuity, and prolonged confusion. The patient with diffusion restriction at the level of splenium CC in magnetic resonance imaging (MRI), whose seizure frequency increased, was evaluated by a neurology doctor. In his electroencephalography, spike waves were observed in both hemispheres, which were frequently generalized. The same lesion was not observed in the diffusion and MRI taken eight days later in the patient who had no seizures in the follow-up.

Conclusion: Studies have shown that CC damage results in the disruption of cortical functions, with disconnection of the cerebral hemispheres and disturbances in consciousness. The clinical spectrum includes a fairly wide symptomatology. RESLES of the CC is an infrequent finding on MRI. Some of these lesions are associated with epileptic seizures, the sudden withdrawal of the antiepileptic drug, or usage. RESLES, which we reviewed with etiology and clinical findings, still remains a mystery. It will be clarified with wide-ranging studies.

Keywords: Reversible Splenial Corpus Callosum Lesion, epilepsy, carbamazepine.

Introduction

Reversible splenial lesion syndrome (RESLES) at corpus callosum (CC) has been defined in many clinical conditions for example malignancies, infections, metabolic disturbances, etc. Many neurological disorders are associated with lesions affecting the splenium of the corpus callosum (SCC). The spectrum of symptoms is broad and clinical presentations may be indistinguishable. Although the etiopathogenesis of transient focal lesions in the corpus callosum splenium has not been clarified, many theories remain on the agenda, especially in epilepsy patients[1,2].

Case presentation

A 38-year-old male patient with idiopathic generalized epilepsy was admitted to the emergency department with complaints of frequent seizures. After the last seizure, he had headaches, nausea, vomiting, and decreased visual acuity, as well as confusion prolonged. In the history of the patient with generalized epilepsy; It was learned that he had generalized tonic-clonic seizures since the age of eight and that he was under antiepileptic treatment, but he did not comply the treatment. Our patient had been using carbamazepine 1200mg/day since 2019. It was reported by his relatives that he had stopped carbamazepine treatment for the last three days. In his neurological examination, there was a prolonged postictal confusion state. Vital signs were within normal limits. No abnormal findings were detected in the biochemistry and hemogram tests, and the patient's infection parameters were evaluated as normal. Diffusion magnetic resonance imaging (MRI) was performed in the emergency department. The patient with diffusion restriction at the SCC level (Figure 1) was seen by the neurologist

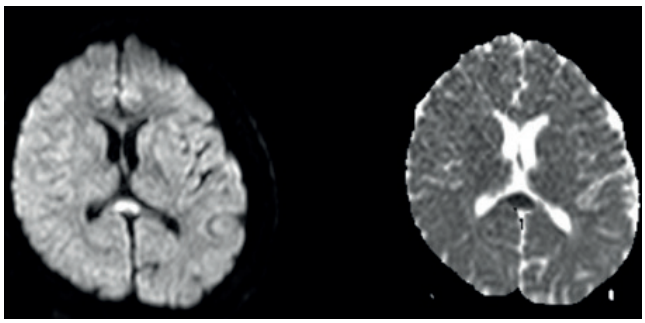

Figure 1:

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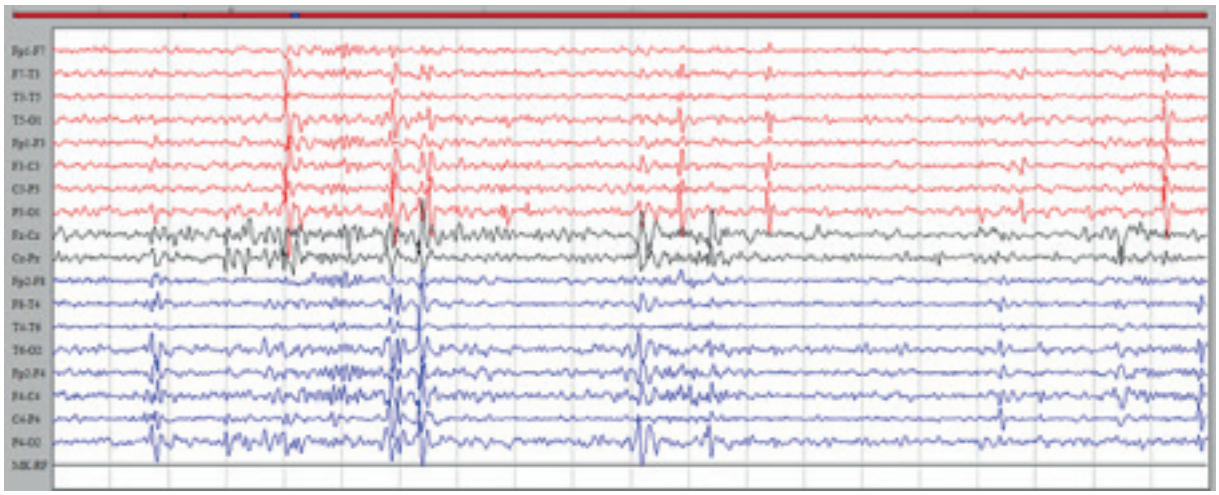

Figure 2:

with a preliminary diagnosis of ischemic stroke. In his electroencephalography, spike waves were observed in both hemispheres, which were frequently generalized (Figure 2). Antiepileptic treatment with levetiracetam 1000 mg/day was started. The seizure of the patient, who was followed up in the neurology service, did not recur and his neurological examination was evaluated as normal two days later. The same lesion was not observed in the diffusion and brain MRI taken eight days later in the patient who had no seizures in the follow-up (Figure 3). In this case, CC transient lesions will be reviewed together with the literature.

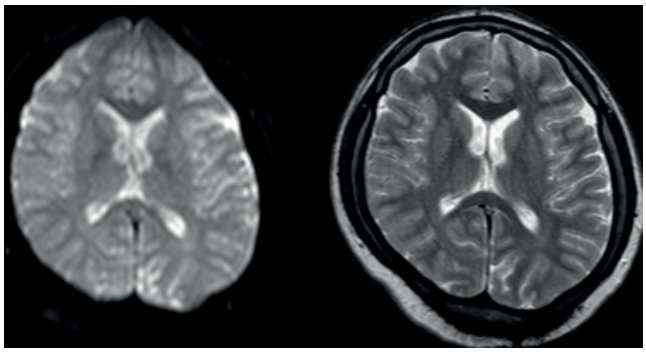

Figure 3:

Discussion

The CC plays a vital role in brain function. Studies have shown that its damage results in the disruption of cortical functions, with disconnection of the cerebral hemispheres and disturbances in consciousness[3]. There have been various terms used to describe splenial lesions; mild encephalitis with a reversible lesion in the splenium (MERS), RESLES, and cytotoxic lesions of the corpus callosum (CLOCCs). MERS is an acute cliniconeuroradiological syndrome characterized by mild encephalopathy or encephalitis presenting as a reversible solitary mass in the central portion of the SCC[4]. However, encephalitis/encephalopathy is not always mild. The spectrum of RESLES includes MERS. CLOCCs are various entities associated with a variety of causes with restricted diffusion, and some of these lesions are irreversible. The splenial lesion of MERS and RESLES contains two different patterns according to the lesion location: Type 1, an isolated lesion located in the center of the splenium of CC, mostly round or oval, with some of the lesion extended along the splenium; type 2, a lesion centered in the splenium and extended into other brain areas[4-7]. Our case of RESLES was all isolated symmetrical lesions in SCC without any extracallosal lesions. Most of the previous descriptions of RESLES have been in the case report format; thus, the incidence of RESLES remains unknown[8].

RESLES of the CC is an infrequent finding on MRI. Some of these lesions are associated with epileptic seizures, the sudden withdrawal of the antiepileptic drug (AED), or usage. While different hypotheses have been generated, its pathophysiology is still not well understood. The possible mechanisms for the restricted diffusion of the SCC include intramyelinic edema, reversible demyelination, damage to the blood-brain barrier, arginine vasopressin release, and inflammatory cell-induced cytotoxic edema[8-10]. Antiepileptic drug toxicity and associated changes in salt homeostasis and transhemispheric seizure propagation are other suspected mechanisms[11,12]. Treatment with antiepileptic drugs like carbamazepine and the rapid concentration changes of drugs can influence fluid balance systems through arginine vasopressin release. These drugs can also increase the number of pro-inflammatory and pro-convulsive cytokines. Öztoprak et al. proposed a possible mechanism of onset in RESLES for patients experiencing seizures. The authors suggest that the discharge of the corpus callosum disseminated in seizures caused a decrease in free water dispersion in the CC. Further studies are needed to investigate this mechanism[8,13]. A lesion of the SCC in patients with epilepsy might be induced by a rapid and relatively long-lasting reduction of antiepileptic drugs and is not associated with toxic drug effects or high seizure frequency[10].

Clinical symptoms included headaches, amnesia, confused consciousness without seizure, nausea, vomiting, and diarrhea, fever, dizziness, visual disturbances, and sensorimotor hemiparesis, dysmetria, dysarthria. The heterogeneity in the clinical manifestations makes RESLES hard to predict before MRI. We present an epileptic patient who was found to have an isolated, reversible lesion in the SCC associated with the sudden withdrawal of carbamazepine. Reversible SCC lesions were also reported in non-epileptic patients using other AEDs[14,15]. In the literature, transient corpus callosum splenium lesions caused by the reduction of levetiracetam, in addition to carbamazepine, have been presented. On the other hand, temporary corpus splenium lesion caused by decreasing antiepileptic doses in epilepsy patients under mono or polytherapy has been reported in many case reports[11,16].

In the case series in which 23 patients with diffusion restriction in the CC were examined, a reversible lesion had observed in 10 patients, and an oval or round lesion with a central location in the CC was detected, similar to our case. In this case series, a symmetrical boomerang-shaped lesion was observed in marchiafava bigami syndrome, while eccentric, irregular lesions had observed in SCC in stroke patients[17].

RESLES has a benign prognosis and is usually associated with complete recovery without any obvious neurological sequelae shortly after the acute course, as in our patient.

Conclusion

RESLES, which we briefly reviewed with their extensive etiology and clinical findings, still remains a mystery. These lesions will be more understandable with new case series and long-term follow-up.

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