A case of epilepsy partialis continua of abdominal muscles after brain tumor surgery

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Abstract. – Epilepsia partialis continua (EPC) is a rare form of focal motor status epilepticus characterized by continuous muscular twitches or jerks involving a limited part of the body, usually facial region and distal limb. Although the cerebrovascular disease is known to be one of the most common causes of this condition, other reported cases with predominant abdominal involvement have different aetiologies, including, tumors, focal cortical dysplasia, and central nervous system infections. No cases of epilepsy partialis continua of the abdominal wall occurred after brain surgery have been previously reported. We describe the clinical, electrophysiological, and neuroimaging findings in an adult patient presenting with persistent unilateral abdominal myoclonus configuring an EPC as the evolution of a super-refractory hemibody convulsive status epilepticus, occurred after brain tumor surgery.

Key Words: Epilepsy, Epilepsia partialis continua, Status epilepticus, Brain surgery.

Introduction

Epilepsia partialis continua (EPC) is a rare form of focal motor status epilepticus that is characterized by continuous muscular twitches or jerks of a limited part of the body, lasting from hours to weeks or years1-2. It typically involves facial region and distal limb, although trunk or abdomen may also rarely be affected2-5. Cerebrovascular lesions have been referred to be one of the common causes of EPC in adults4. However, in cases of abdominal involvement, most documented causes differ and primarily include: brain tumors or metastasis5-6, focal cortical dysplasia1, central nervous system infections6, and subdural haematoma7. In general, unilateral or bilateral contractions of the abdominal muscles are an uncommon manifestation of the epileptic seizures. The pathophysiology of this peculiar and rarely encountered ictal clinical semiology is still not completely defined and the anatomic localization of the epileptogenic zone in these seizures has been debated, thus reflecting the complex cortical organization and the somatotopic representation of abdominal musculature. However, circumscribed frontal parasagittal or parietal lesions have been well-documented causes of truncal seizures5,6,8. We describe the electro-clinical and neuroimaging features of an adult patient presenting with persistent clonic twitching of the abdominal muscles that were considered to represent a rare manifestation of EPC as the evolution of a super-refractory hemibody convulsive status epilepticus occurred after brain tumor surgery.

Case Description

A 61-year-old man with an unremarkable past medical history was admitted to the Intensive Care Unit (ICU) for a 48-hours history of continuous motor jerks involving left hemibody with impaired awareness. Approximately one month before, he undergone a right-sided craniotomy for the elective resection of a temporal lobe mass lesion (Figure 1A) heralded by a non-motor focal seizure. The postoperative course had been regular and neither focal neurological deficits nor seizures had been observed. The histopathological examination had revealed a solitary fibrous tumor/hemangiopericytoma (grade II WHO). At the ICU admission, the patient was intubated, with continuous intravenous midazolam (cIV-MDZ) infusion at anesthetic dose. The vital signs were stable. Left side forced eyes and head deviation, as well as sub-continuous motor jerks involving left hemibody, were present. A brain MRI scan showed the tumor resections’ cav-
ity in the right temporal region (Figure 1B); diffusion-weighted imaging (DWI) sequences (Figure 1C) revealed a focal restricted signal, mainly involving the right parietal region. Continuous EEG monitoring, which showed a sustained epileptic activity involving all contacts over the right hemisphere, confirmed a condition of super-refractory focal convulsive status epilepticus. Due to its inefficacy, cIV-MDZ was discontinued and a general anesthesia was started and maintained for approximately 24 hours by using Propofol infusion at a dose that results in EEG burst suppression. Within 3 hours of discontinuing Propofol infusion, the patient recovered consciousness; focal motor seizures of left hemibody were no longer observed. A slight Todd’s hemiparesis was evident, as well as irregular, variable, hyperkinetic paroxysmal movements of the abdominal wall. Intermittent vigorous and pseudo rhythmic contractions of the abdominal muscles markedly more intense on the left side. Note that the muscle contraction produces a diffuse movement of the abdominal wall, predominantly on the left side. The concomitant EEG activity was characterized by rhythmic epileptiform activity involving all contacts over the right hemisphere (Figure 1D). Oral clonazepam (1 mg t.i.d) was then added to existing treatment with levetiracetam (1000 mg t.i.d.) and carbamazepine (200 mg t.i.d.). In the following days, the patient experienced a progressive clinical improvement with subsequent complete disappearance of the abdom-

Figure 1. A, Brain MRI scan (T1-weighted post gadolinium coronal and axial images), showing a right temporal lobe mass lesion characterized by a cystic component, contrast-enhancement and regional edema. B, Brain MRI scan (T2-weighted fluid-attenuated inversion recovery coronal and axial images), showing the tumor resection cavity in the right temporal region. C, Brain MRI scan diffusion-weighted imaging (DWI) sequences, acquired during sustained epileptic activity, showing a focal restricted signal mainly involving the right frontal and parietal cortex. D, EEG showing a mild slowing of background activity and epileptiform activity. This manifests as rhythmic spikes and spike-wave discharges involving right frontal and centro-parietal areas with early spreading over the temporal regions. E, Follow-up DWI maps with no areas of diffusion restriction.
inal myoclonus. At one-month follow-up visit, he did not complain about further neurological symptoms; brain MRI showed disappearance of DWI alterations and was unremarkable for new lesions (Figure 1E). Standard EEG documented a normal background activity and interictal brief bursts of slow waves over the right temporal region.

Discussion

The case we are reporting is representative of the ECP that exclusively involved the abdominal muscles as an evolution of a focal motor super-refractory SE, affecting the entire left side of the body, and occurring after brain surgery. This case appears worth of note both for semeiological and aetiological aspects. Indeed, isolated clonic movements of the abdominal wall are unusual manifestations of EPC, that usually involve distal limb or facial muscles according to their wide representation in primary motor cortex. In general, the involvement of the abdominal musculature as a clinical manifestation of a focal motor seizure is rarely observed, both for anatomical and physiopathological reasons. Firstly, the trunk is well-known to have a small topographic representation on the motor cortex. Secondly, the epileptic threshold of the cortical area of trunk representation is considered to be higher than other primary motor areas and, as a consequence, such clinical manifestation may not be evident during seizure activity. Several cortical regions have been reported to be associated to epileptogenic lesions in cases of EPC involving abdominal musculatures, such as the parietal lobe, frontal lobe, and parasagittal areas. None of these areas were restricted to the somatotopic representation of abdominal musculature. The notion of multiple localizations associated with a common clinical manifestation of abdominal contractions may be justified by the complex organization of the homunculus and some individual variability. However, according to general concepts on presurgical evaluation in epilepsy surgery, it is well-established that the symptomatogenic zone could not necessarily coincide with the epileptogenic lesion. As far aetiological aspects are concerned, the ECP of the abdominal wall has been previously reported in other different neurological conditions, including cerebrovascular lesions, brain tumors or metastasis, focal cortical dysplasia, central nervous system infections and subdural haematoma.

To the best of our knowledge, no other cases of EPC of the abdominal wall after brain surgery has been reported. Seizures related to craniotomy may occur even not acutely (immediate or early, within 24 hours or 1 week, respectively), but also after a silent period (late, after two or more weeks). Two different, but closely linked, mechanisms have been hypothesized to explain the pathogenesis of these epileptic events: extravascular blood leakage generating free radicals and ischemic/hypoxic processes leading to cell trans-membrane ion imbalance and, consequently, to an increased regional or diffuse cortical hyperexcitability. Although in the present case the epileptic nature of the clinical phenomenon was overt for clinical presentation as an evolution of a super-refractory hemibody convulsive status epilepticus, continuous movements of the abdominal wall may also be related to other neurological conditions, such as belly dancer’s dyskinesia, incoercible hiccups or propriospinal myoclonus. Consequently, the role of EEG monitoring has been confirmed to be crucial in contributing to differential diagnosis and therapeutic management of these conditions.

Conclusions

EPC with isolated involvement of the abdominal muscles, often associated to structural lesions involving different cortical regions, has been rarely reported. According to classical principles, the abdomen or trunk has a limited representation area on the cortical homunculus. Nevertheless, brain surgery or other structural conditions may lead to a reorganization of cortical substrates and networks, thus they give rise to spreading discharges through a distant part of homunculus or propagation of discharges through to the deep layers instead of the surface.

Conflict of Interest

The Authors declare that they have no conflict of interest.

References


