Paroxysmal pain as the only presentation of focal epilepsy

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Abstract

A 55-year-old man with left temporo-insulo-parietal glioblastoma was admitted because of abrupt excruciating pain in right arm and ipsilateral face. EEG showed a left central frontal rhythmic activity and complains were controlled with anti-epileptic drugs. Paroxysmal pain is an unusual manifestation of focal seizures and can be the only symptom.

Key Clinical Message:

- Painful seizures can be the only feature of focal epilepsy.

- Its localization on a limb favors the epileptic etiology while abdominal or cephalic paroxysmal pain is more unspecific.

- Painful somatosensory seizures arise from operculo-insular cortex.

Background:

Ictal pain is a rare manifestation of focal seizures, with a frequency ranging from 0,2 to 2,8% in patients with epilepsy[1]. Classically, it is categorized according to its localization as cephalic, abdominal (visceral or pneumogastric) or unilateral somatosensory pain[2]. The last is usually striking and can be described as a burning, stabbing, prickling, throbbing, or tearing sensation. In most cases they are accompanied by other motor, sensory or behavioral features which can denote their ictal origin[3]. When it occurs in isolation, seizures are often misdiagnosed and patients can go through unnecessary diagnostic procedures and inadequate treatment. While most patients who present with paroxysmal headache or abdominal pain do not have an epileptic cause in their origin, the localization of paroxysmal pain on a limb is more suggestive of an underlying epileptic cause[1].

Case Presentation:

A 55-year-old man was diagnosed with a left temporo-insulo-parietal left tumor after developing focal cognitive seizures with aphasia and complex visual hallucinations (fig. 1). He underwent partial tumor resection resulting in persistent dysphasia. The histopathology diagnosis was compatible with a glioblastoma. He was medicated with levetiracetam and sodium valproate due to increasing of seizure frequency, characterized by transitory worsening of previous dysphasia. He started treatment with chemotherapy and radiotherapy (according to STUPP protocol), with an initial good radiological response. While undergoing oral chemotherapy cycles, he was admitted in the Emergency Room, desperately screaming due to acute and excruciating pain on the right arm and ipsilateral face, described as shock-like and stabbing. Each episode lasted a few seconds but they had been repeating in cluster for over an hour. General blood analyses were unremarkable and the electroencephalography showed a left central frontal rhythmic activity during the pain episodes (fig. 2). The add-on of intravenous lacosamide resolved the painful complaint in a few minutes. The brain MRI performed a few days later showed local tumor recurrence (fig. 2).

Discussion:

Although ictal pain is often associated with other seizure symptoms, it can be the only manifestation of focal epilepsy and differentiating them from others can be challenging. Among patients with ictal pain, those who report it to their limbs are more likely to have an epileptic cause[1].

Under some pathophysiological conditions, operculo-insular lesions may produce location-specific painful epileptic seizures. Functional imaging studies have identified several cortical areas activated by painful stimuli, referred as "pain matrix", including, among others, the primary somatosensory area, the supplementary motor area, the insula, the anterior frontal or the posterior parietal cortices[4]. Some cases of ictal pain with foci originating in the frontal, parietal and temporal regions have been described with surface EEG[3,5]. However, data from functional cortical mapping by using direct cortical stimulation suggests that the origin of painful somatosensory seizures arises solely from the medial part of parietal operculum or the posterior and upper part of the insular cortex[6,7].

Taking into account the cortical mapping studies and the localization of our patient's glioblastoma, we suggest that the painful seizure has a left operculo-insular origin and that the rhythmic activity captured on the surface EEG represents propagation to the primary parietal cortex.

Paroxysmal contralateral pain can be the only manifestation of epileptic seizures with operculo-insular cortex involvement. This knowledge is useful to avoid misdiagnosis and to prompt appropriate management with antiepileptic drugs.

Author Contribution Statement:

DG: wrote the manuscript; MF and JM: evaluated the patient and revised the manuscript; JF: carried out the EEG assessment. All authors approved the final version to be published.

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