Epileptic Disord 2017; 19 (2): 207-11

Focal epilepsy with paroxysmal pain due to somatic injury

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Received July 16, 2016; Accepted April 18, 2017

ABSTRACT – We present two cases with paroxysmal pain that developed after a somatic injury to the trunk. The main characteristic of the episodes was paroxysmal severe pain, mainly located in the original region of somatic injury, with ipsilateral tonic or dystonic behaviour. The clinical characteristics supported a diagnosis of focal epilepsy. Both scalp EEG and MEG findings suggested epileptic activities on the contralateral central cortex. The focal seizures had a good response to antiepileptic drugs. It is hypothesized that peripheral somatic injuries can modify cortical excitability and lead to plastic changes in the sensory/motor cortex, ultimately resulting in focal seizures. We provide additional evidence for the phenomenon that a peripheral somatic injury could induce focal epilepsy. [*Published with video sequence on www.epilepticdisorders.com*]

Key words: seizure, pain, peripheral injury, epilepsy

VIDEO ONLINE

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doi:10.1684/epd.2017.0918

Few reports are available that describe focal epilepsy developing after a peripheral somatic injury (Spiller *et al*., 2005; Sumitani *et al*., 2011). Furthermore, this kind of focal seizure is rarely characterized by paroxysmal pain, similar to the sensation at the instant of injury. In this article, we present two cases of focal epilepsy with paroxysmal pain that developed after a somatic injury to the trunk. The main characteristic of the episodes was paroxysmal severe pain, mainly located in the region of original somatic injury, with ipsilateral tonic or dystonic movement. We hypothesize that some plasticity of the neural circuit was activated accidently by the somatic injury, and this activation was responsible for the following seizures.

Case series

Patient 1

A 33-year-old woman with no significant medical or family history was referred for investigation of pain and abnormal movement. Ten months ago, the patient was injured by a hard object on her left rib, causing moderate pain in the left part of her chest. One week later, her pain clearly developed with paroxysmal tearing-like or electric

shock-like feelings. The pain was accompanied by stiffness and tonic jerks of the left limbs. During the episodes, the patient felt that she could not move her left limbs and had shortness of breath (*see video sequence*). Sometimes, she fell down to the right side. These symptoms lasted for several seconds and gradually increased to dozens of times per day. In the interictal period, she did not feel any pain or stiffness. She was initially diagnosed with intercostal neuralgia and was prescribed voltaren and gabapentin, which had a poor effect.

At the time of admission, her interictal neurological examination was normal. The location of the paroxysmal pain is marked in *figure 1*. An abdominal ultrasound and chest CT scan were normal. Brain MRI indicated mild cerebral atrophy in the bilateral parietal lobes. An interictal scalp EEG showed sharp slow waves in the right central and parietal regions. Rhythmic slow activities during the ictal phases could be observed in the same regions (*figure 1*). The MEG examination (Elekta Neuromag Vector View 306 Channel Meg) suggested that the interictal spike dipoles were located in the right parietal lobe (*figure 1, 2*). Carbamazepine (0.1 g every 12 hours) was then administered and the seizure frequency was reduced dramatically to 1-3 times per week. Surprisingly, the pain was also relieved significantly.

Patient 2

A 19-year-old man with no significant medical or family history was admitted for assessment of pain and epilepsy. Ten years earlier, he had a right clavicular fracture due to a trauma, causing severe focal shoulder pain. After internal fixation, the pain was relieved temporarily. Five years ago, the patient felt severe tear-like

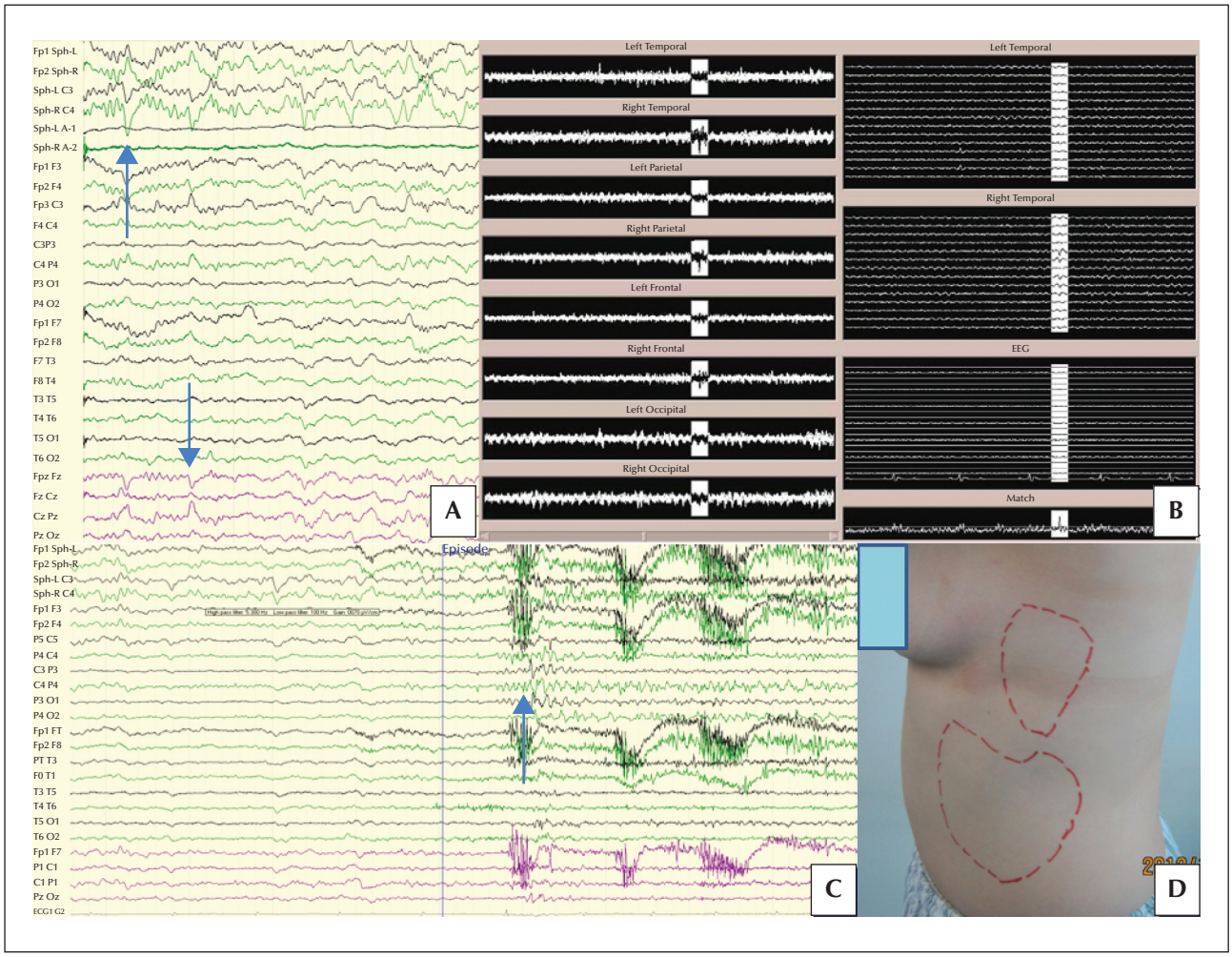

Figure 1. The location of paroxysmal pain and abnormalities on EEG and MEG in Patient 1. (A) Interictal sharp slow wave in the right central-parietal region on scalp EEG (arrows). (B) Epileptiform MEG discharges in the right parietal and temporal region on MEG (goodness of fitness: 91.2%). (C) Ictal rhythmic activities originated from the right central and parietal region on scalp EEG (arrow). (D) The location of the paroxysmal pain in the trunk is marked.

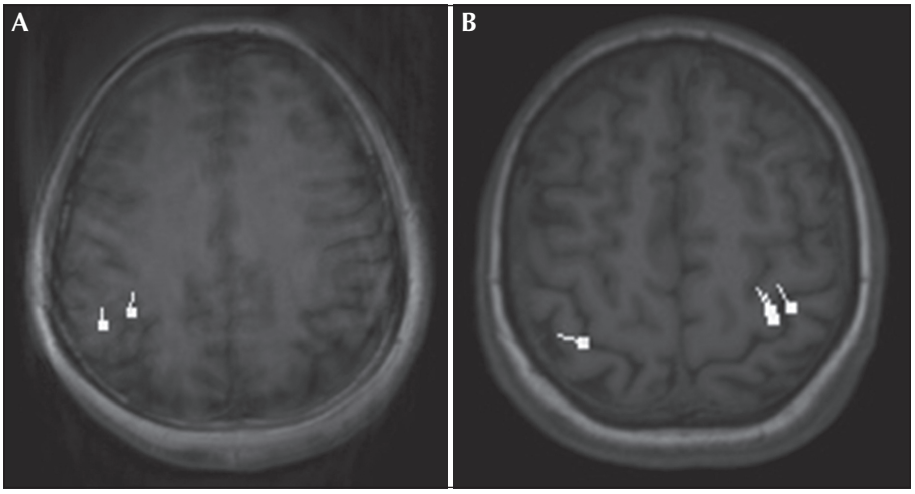

Figure 2. The interictal spike dipoles on MEG of the two patients. The spike dipoles on MEG were located in the right parietal lobe in Patient 1 (A) and in the bilateral parietal lobe, dominantly on the left side, in Patient 2 (B).

pain in his right shoulder intermittently, and he felt that his right arm was twisted to his back during the paroxysmal pain. These symptoms lasted several seconds and occurred 2-3 times per month. During the episodes, he could sometimes control the progression of pain by an active hard movement of his right shoulder and arm. In contrast, in the interictal period, sometimes the symptoms could be induced by a hard movement of the right shoulder.

At the time of admission, his interictal neurological examination was normal. Brain MRI was normal. A MEG examination (Elekta Neuromag Vector View 306 Channel Meg) indicated that interictal spike dipoles were located in the bilateral parietal lobe, dominantly in the left side (*figure 2*). An interictal scalp EEG showed sharp slow waves in the left central and parietal regions. Levetiracetam (0.5 g every 12 hours) was given and following this, seizures rarely occurred.

Discussion

The fact that a peripheral somatic injury could induce focal epilepsy is an interesting phenomenon. Only a few patients have been reported to have focal epilepsy which developed after a peripheral somatic injury (Spiller *et al*., 2005; Sumitani *et al*., 2011). In this report, we have tried to provide additional evidence for this phenomenon by investigating two cases, who developed focal epilepsy with paroxysmal pain due to a somatic injury. In contrast to the former reports in which the patients hurt their limbs, our two patients had peripheral injuries to the trunk. These two patients suffered from focal paroxysmal attacks, which occurred one week and five years, respectively, after the somatic injuries. During the episodes, these two patients felt tear-like pain in the injury area, followed by tonic or dystonic movement in the ipsilateral trunk or limb.

Above all, we firstly sought to investigate whether the episodes of the two patients were epileptic seizures. Although there were no secondary generalized seizures, some clinical characteristics of the episodes supported a diagnosis of epileptic seizure. The paroxysmal pain accompanied by tonic/clonic stiffening of focal muscles occurred repetitively. The part of the body involved was more extensive during the seizure relative to the somatic injuries. The pain was not distributed along the peripheral nerve, such as the intercostal nerve or brachial plexus. There was no skin damage, no abnormality of skin temperature, and no hypalgesia or hyperpathia. Therefore, we can reasonably exclude a diagnosis of neuralgia or complex regional pain syndromes. Although there were occasional motor-evoked seizures in the second patient, most of the seizures occurred spontaneously. The pain and the tonic/clonic behaviour occurred paroxysmally, lasting a short time, whereas there was no pain or any functional disability in the interphase. Although there was no typical spike, the interictal sharp slow waves on the scalp EEG also provided some evidence for epileptic diagnosis. A specific difference from that of the patients of Spiller *et al*. (2005) is that the interictal spike dipoles clustered in the contralateral parietal lobe during the MEG examination. It was reported that MEG is particularly sensitive to tangential currents, which presumably arise preferentially from the cortical sulci, and MEG can offer the advantage of directly localizing neuronal activity in excellent temporal resolution on a scale of milliseconds rather than relying on the accompanying haemodynamic or metabolic changes (Van Veen *et al*., 1997; Nowak *et al*., 2009; Mohseni *et al*., 2012; Mohamed *et al*., 2013). Therefore, the dipoles

on MEG may be further confirmation of focal epilepsy. Additionally, these patients, as well as those of Spiller *et al*. (2005), showed a positive response to antiepileptic drugs, supporting the diagnoses of focal epilepsy. Therefore, most of the evidence supported a diagnosis of epilepsy in these cases, although there remained uncertainties regarding the accuracy of the diagnosis. This therefore raises the interesting question: how could a somatic injury result in focal epilepsy with paroxysmal pain? Spiller *et al*. hypothesized that peripheral somatic injuries can modify the cortical neuronal circuit which leads to plastic changes in the sensory/motor cortex, resulting in abnormally increased excitability. The excitability ultimately increases to the extent that it results in seizures (Spiller *et al*., 2005). Paglioli *et al*. proposed that a peripheral trauma can provoke a massive afferent volley and act as a disinhibiting trigger which breaks the delicate balance between intrinsic hyperexcitability and inhibition in neuronal networks (Paglioli *et al*., 2016).

Some examples of centrales neuronales plasticity and reorganization that result from either peripheral stimulation or injury havent been reported and illustrated by previous authors (Ramachandran, 1993; Druschky *et al*., 2000; Hickmott and Merzenich, 2002; Elbert and Rockstroh, 2004). Striano emphasized the pioneering studies by Amantea on "epilepsy from afferent stimuli" (Striano and Striano, 2006). In these studies, it was found that clonus induced by the application of strychnine to the sensory/motor cortex could be enhanced by the stimulation of the peripheral area, somatotopically related to an excited cortex. A "reflex mechanism" is thought to be likely to be involved in the changes of cortical excitability (Vizioli, 1962; Striano and Striano, 2006). This also helps to explain an additional phenomenon described in Case 2; sometimes, during the interictal period, the pain could be elicited by a hard movement of the right shoulder. Conversely, the pain could also be stopped by an active hard movement of his right shoulder during some episodes. This activity is similar to ligature therapy which had been proven to be effective in appropriate situations for centuries. A peripheral stimulation may excite or inhibit excessive excitability of the focal sensory/motor cortex. These activation and termination phenomena reveal the relationships between sensory/motor circuit and focal epilepsy.

However, another puzzling question is why only very few patients who experience somatic injury suffer from focal epilepsy. Paglioli *et al*. also raised the question whether augmented cortical hyperexcitability following peripheral injury occurs only in the presence of a pre-existing abnormal cortex (Paglioli *et al*., 2016). In their opinion, massive afferent volleys likely disinhibit a pre-existing hyperexcitable cortex (Palmini, 2010). In Patient 1, the abnormal MRI indicating mild cerebral atrophy in the bilateral parietal lobes may be a predisposing factor for epilepsy. In contrast, in Patient 2, there was no visible abnormality on MRI which might account for why there was much more latency regarding the development of seizure after the somatic trauma. Further investigations including positron emission tomography (PET) and ictal single-photon emission computed tomography (SPECT) studies may be helpful to confirm the diagnosis of focal epilepsy and reveal the mechanism of seizure development in such clinical settings. □

Disclosures.
None of the authors have any conflict of interest to declare.

Legend for video sequence

The patient felt paroxysmal pain on her left trunk. Then her left limbs became stiff and tonic for about 20 seconds. She rubbed her left chest to release the pain when the stiffness remitted.

Key words for video research on www.epilepticdisorders.com

Phenomenology: aura (somatomotor), motor seizure (simple), tonic posture
Localisation: central motor, central (right), central sensory
Epilepsy syndrome: not applicable
Aetiology: not applicable

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