

Prolonged ictal monoparesis with parietal Periodic Lateralised Epileptiform Discharges (PLEDs)

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ABSTRACT – We report a patient with prolonged monoparesis and parietal periodic lateralised epileptiform discharges (PLEDs). The patient was a 73-year-old man with chronic myelomonocytic leukaemia who developed persisting monoparesis of the right arm, sensory aphasia, and finger agnosia, initially associated with focal clonic seizures. These neurological deficits remained for seven days without subsequent focal clonic seizures. The EEG showed left-sided PLEDs, maximal in the left occipito-parietal area. Ten days later, following phenytoin treatment, these symptoms suddenly improved and parietal PLEDs disappeared. Sustained PLEDs in the left parietal region may have been causally associated with ictal paresis in this patient.

Key words: inhibitory motor seizure, PLEDs, EEG

Ictal paresis or inhibitory motor seizures are rare but an important clinical condition that should be differentiated from other neurological emergencies such as stroke and acute encephalitis, since it may improve with appropriate antiepileptic treatment. However, the diagnosis is sometimes difficult to make by the bedside alone and there are only a few reports with documented EEG or imaging during actual paresis (Matsumoto *et al.*, 2005; Bussière *et al.*, 2005; Villani *et al.*, 2006; Calarese *et al.*, 2008). Pathophysiology of inhibitory

motor seizures has not been fully elucidated. Periodic lateralised epileptiform discharges (PLEDs) are relatively uncommon EEG findings, though they were well described (Pohlmann-Eden *et al.*, 1996). Although PLEDs appear in some patients with focal seizures, their significance regarding ictogenesis, as well as pathophysiology, remains obscure. Here, we describe a patient with prolonged ictal monoparesis and parietal PLEDs, and discuss the pathophysiological significance of parietal PLEDs in relation to ictal paresis.

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Case report

A 73-year-old man with chronic myelomonocytic leukaemia was admitted to Kyoto University Hospital. He presented with focal clonic seizures of the right arm which spread to the right side of the face (Day 1). He had no previous history of epilepsy, seizures, or other neurological deficits. On examination after the seizures had stopped, he had mild monoparesis of the right hand, finger agnosia, and sensory aphasia. These neurological signs continued over the following several days, as discussed below. Brain MRI revealed a slight chronic subdural haemorrhage (SDH) on the left lateral convexity, but no acute intracerebral lesion at all (*figure 1*). Serum biochemical testing did not show any severe metabolic disorder, including hypoglycaemia, uraemia or hepatic insufficiency, which could be responsible for focal neurological deficits.

The scalp EEG showed left-sided PLEDs, maximal in the left occipito-parietal area (*figure 2A*) throughout the entire 30-minute recording. Rhythmic 3-4Hz activity developed in the left frontal region, approximately one second before the convulsion of the right hand (*figure 3*). These right hand convulsive seizures occurred twice during the EEG recording on Day 1. The patient was given a standard dose of intravenous phenytoin (PHT). Although brief, focal, clonic seizures of the right hand occurred on Day 2 and 3, the mild monoparesis of the right hand, finger agnosia, and sensory aphasia remained constant. Immediately before and after seizures, the degree of right hemiparesis did not change significantly. The EEG recording on Day 5 revealed continuous, right, parietal PLEDs, which were similar to those on Day 1. Based on these findings, the patient was diagnosed with right hand focal motor seizures, and the persisting monoparesis was most likely to have resulted from ictal paresis associated with PLEDs, rather than Todd's paresis, either alone or in addition. Administration of intravenous PHT was continued. He was observed closely in the critical care room and underwent continuous cardio-respiratory monitoring. Afterwards, brief, focal, clonic seizures of the right hand occurred on Day 10. Nine days after starting PHT (Day 10), sustained monoparesis of the right hand, finger agnosia, and sensory aphasia suddenly improved and parietal PLEDs disappeared (*figure 2B*).

Discussion

In our patient, persisting monoparesis of the right hand, finger agnosia, and sensory aphasia were most likely ictal manifestations associated with parietal PLEDs, since there was no causative intra-parenchymal lesion on brain MRI or CT, and these symptoms quickly

resolved once EEG improved with PHT. As an alternative explanation, Todd's paralysis or postictal transient paralysis following partial seizures may be considered, either occurring alone or in addition. However, the present condition may not be explained by Todd's paralysis alone because: (1) these symptoms persisted for seven days after the development of clonic seizures; (2) brief, clonic convulsion of the right upper extremity (positive motor event) occurred alongside sustained monoparesis of the same limb (negative motor event); (3) the degree of paresis did not essentially change before and after brief convulsive seizures; and (4) paresis quickly recovered when PLEDs disappeared. Nevertheless, the possibility that an effect of Todd's paralysis might have partially contributed to persistent monoparesis cannot be excluded. The chronic SDH may be related to the epileptic focus present as well as the PLEDs.

Ictal paresis or inhibitory motor seizures are a rare clinical condition, and from a clinical point of view, importantly, should be distinguished from other situations such as cerebral ischaemia, brain tumour or acute encephalitis. However, the pathophysiology of inhibitory motor seizures has not been fully elucidated because there are only a few available reports of documented EEG or imaging during actual paresis.

Our group has previously reported three patients with ictal arm monoparesis (Matsumoto *et al.*, 2005). In these patients, ictal epileptiform discharges were observed in the centro-parietal area and a well-circumscribed lesion was commonly present in the primary arm somatosensory area (S1). Recently, inhibitory motor seizures with epileptic discharges from the contralateral centro-parietal region (Bussi re *et al.*, 2005), temporo-parietal region (Tseng *et al.*, 2006), centro-frontal region (Villani *et al.*, 2006), and fronto-temporal region (Calarese *et al.*, 2008) have been reported. These reports support the notion that cortical areas responsible for inhibitory motor seizure are located *in* or *close* to the perirolandic area. Some previous studies have revealed that motor responses of the primary hand motor area evoked by electrical stimulation were widely distributed in both precentral and postcentral gyri (Nii *et al.*, 1996; Fukuda *et al.*, 2008). Nii *et al.* (1996) also showed that inhibitory motor responses were precipitated by electrical stimuli to the perirolandic area with wide distribution, including pre-motor, primary motor, and primary sensory cortices. In the present patient, the presence of PLEDs in the left parietal region during persisting monoparesis of the right hand strongly suggests that the contralateral parietal area, which includes S1, played an important role in focal inhibitory motor seizures, and furthermore, our patient also developed sensory aphasia and finger agnosia, all of which resolved after the disappearance of PLEDs.

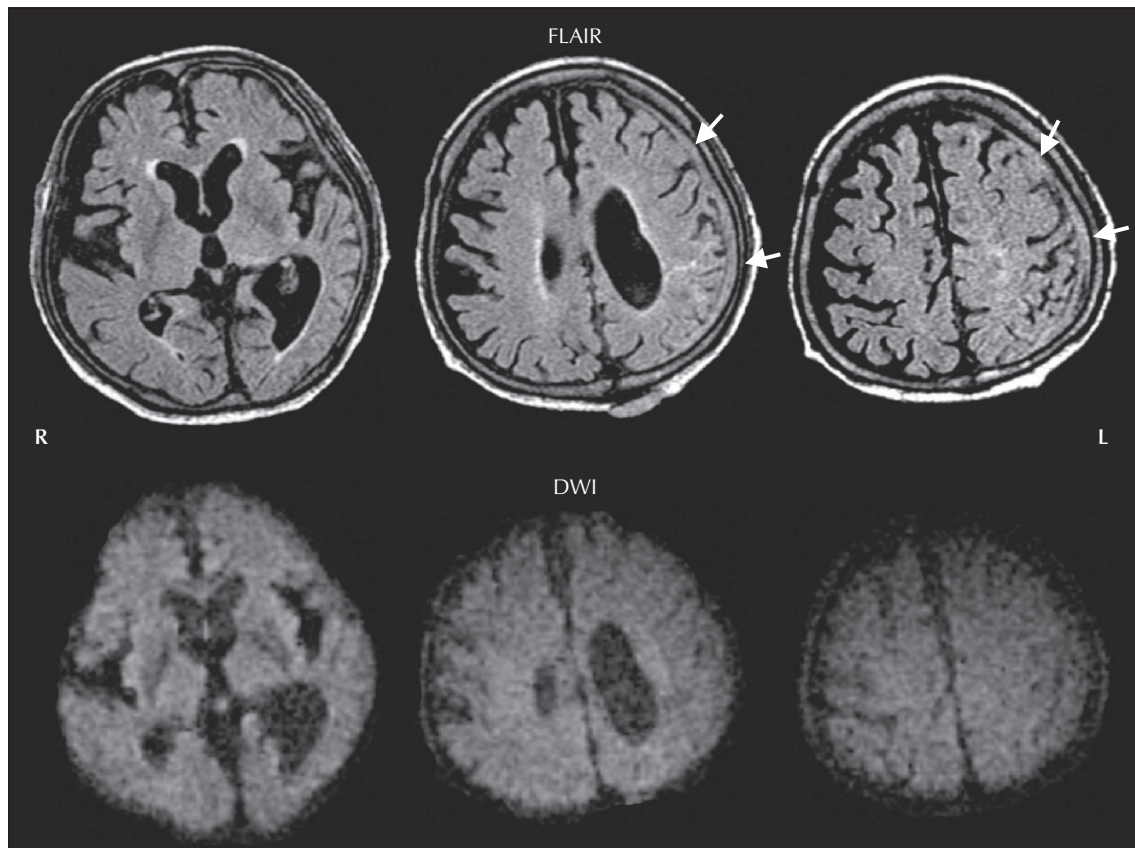


Figure 1. Brain MRI (Day 2) Fluid attenuation inversion recovery (FLAIR) images show a mild degree of subdural haematoma in the left parietal region (upper panel; arrows). No intracerebral abnormal intensity was detected by FLAIR or diffusion-weighted images (DWI; lower panel).

As an ictal manifestation of PLEDs, *epilepsia partialis continua* or repetitive focal motor seizure is common (Pohlmann-Eden *et al.*, 1996). Several studies have described ictal paresis with PLEDs (Bussi re *et al.*, 2005; Tseng *et al.*, 2006; Calarese *et al.*, 2008). Bussi re *et al.* (2005) reported a patient who suddenly presented with weakness of the left side of the face and upper extremity and PLEDs in the right hemisphere, with maximal expression in the centro-parietal and inferior frontal regions. Tseng *et al.* (2006) documented a case of ictal left hemiparesis with PLEDs on the right temporo-parietal region. In these cases, SPECT demonstrated increased perfusion in the same region as the location of maximal PLEDs. However, the degree of temporal consistency between the hyper-perfusion and PLEDs in these cases was not ascertained because the EEG and SPECT were performed on different days. The patients recovered full movement after antiepileptic drug therapy (Bussi re *et al.*, 2005). Calarese *et al.* (2008) described two children with ictal paresis associated with transient contralateral fronto-temporal PLEDs using video-EEG monitoring. In the

present patient, regarding inhibitory motor seizures, PLEDs were an ictal epileptic discharge arising maximally from the left parietal area. It is noteworthy that the patient also presented with several episodes of right hand clonic seizures during a period of persisting right hand weakness with PLEDs. The EEG showed right frontal ictal activity with clonic convulsions, presumably spreading from the right parietal area. Therefore, persisting monoparesis with PLEDs could be regarded as a rather stable, ictal state which could easily provoke frontal, ictal spreading. In this sense, although the ictogenesis of PLEDs remains unresolved, PLEDs may manifest with positive (*epilepsia partialis continua*) and negative motor events (persistent monoparesis), both of which are relatively stable events, but also potentially ictal phenomena. This is consistent with the previous view of PLEDs, reflecting an "ictal-interictal continuum" (Pohlmann-Eden *et al.*, 1996).

As mentioned above, there are several limitations to the interpretation of this case study. We can not completely exclude the possibility of prolonged Todd's

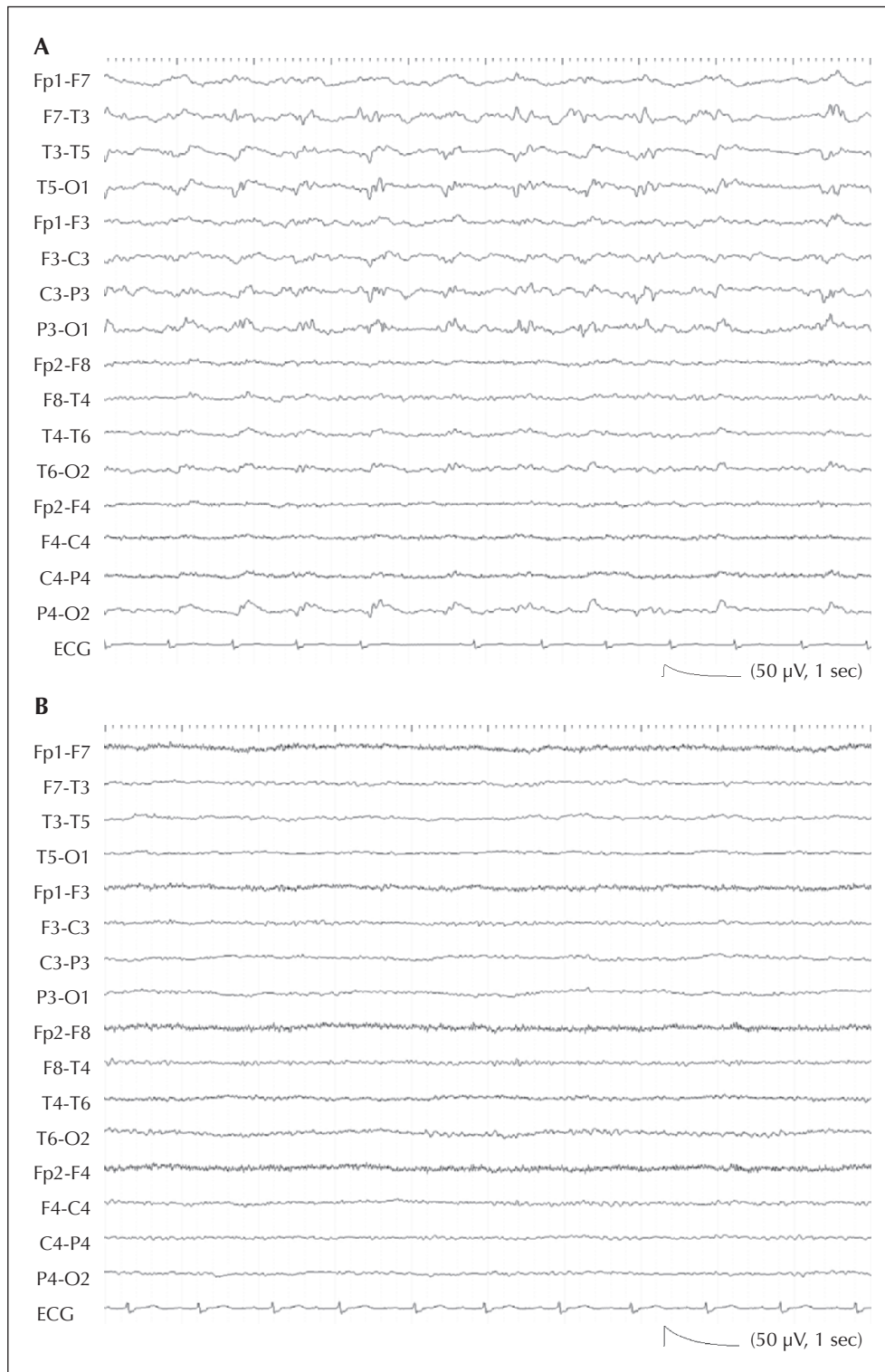


Figure 2. Scalp EEG. (A) Day 2. Scalp EEG shows left-sided PLEDs maximal in the left occipito-parietal region during monoparesis of the right hand, sensory aphasia, and finger agnosia. (B) Day 10. Parietal PLEDs disappeared after monoparesis improved and EEG was almost normal. Fast activity is well preserved in the left posterior quadrant. This suggests an absence of any structural abnormality and presence of only epileptic abnormality during the occurrence of PLEDs.

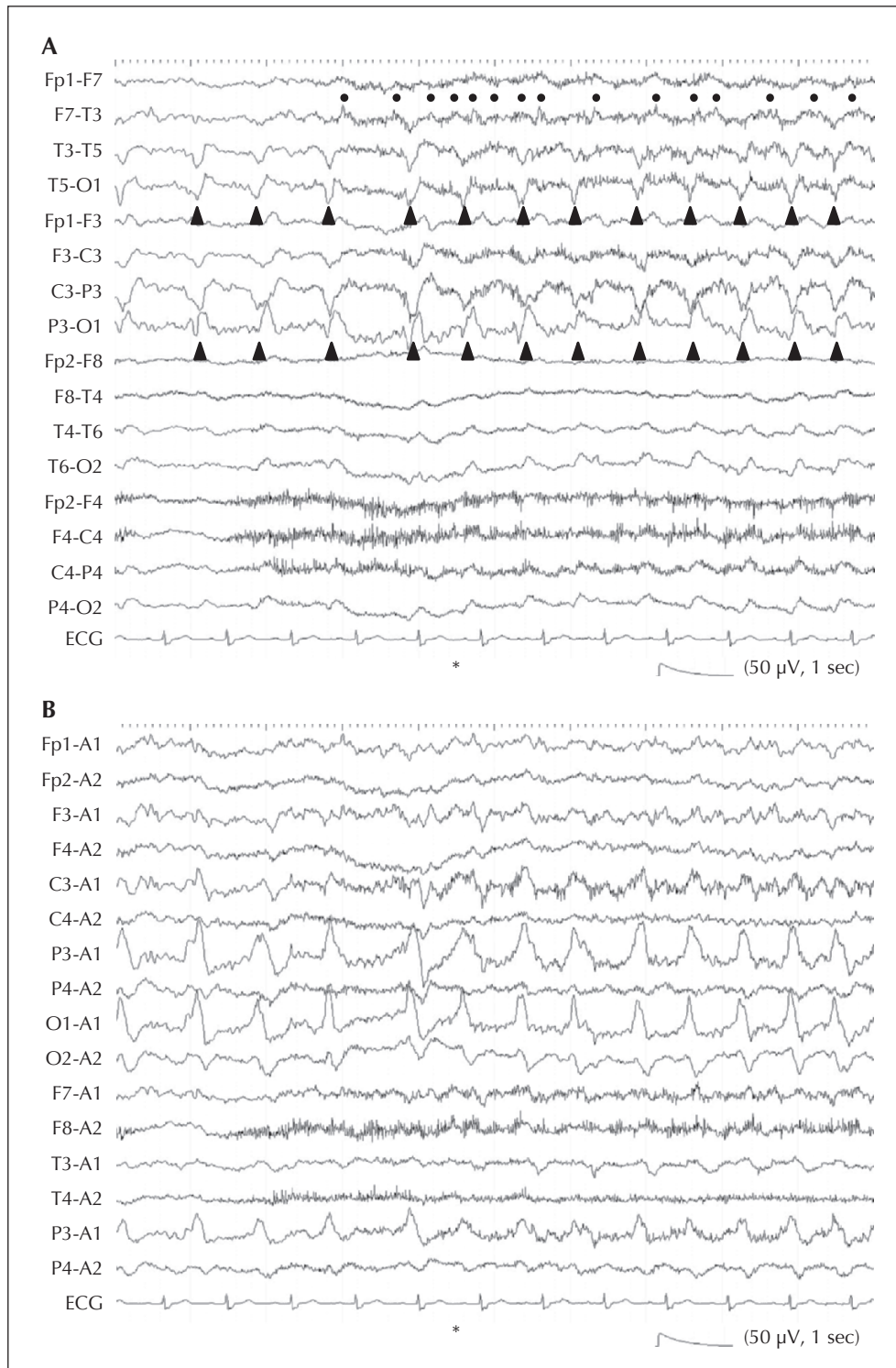


Figure 3. Scalp EEG at onset of tonic convulsion of the right upper limb. A) longitudinal bipolar montage; B) referential montage of the same page. Before the tonic convulsion of the right upper limb appeared, the EEG showed left-sided PLEDs maximal in the left occipito-parietal region (indicated by arrowheads). Immediately before the onset of tonic convulsion (asterisk), the rhythmic 3-4-Hz activity developed in the left frontal region (indicated by dots).

paralysis, because it is impossible to clarify the precise temporal relationship between cessation of the parietal PLEDs and the disappearance of monoparesis of the right hand. Although continuous video-EEG monitoring is essential to document both the improvement of neurological signs and disappearance of PLEDs as precisely as possible, this was not available in this patient. However, it is certain that left parietal PLEDs were closely associated with monoparesis of the right hand, consistent with the literature. Based on the present case report, we would like to stress that even in the case of persisting paresis over a period of days, ictal paresis should be carefully considered clinically.

Ictal persistent monoparesis is one of the significant and treatable conditions that should be included in the differential diagnosis of persistent focal paresis, and PLEDs in the parietal area (including S1) may be associated with this condition. □

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References

- Bussi re M, Pelz D, Reid RH, Young GB. Prolonged deficits after focal inhibitory seizures. *Neurocrit Care* 2005; 2: 29-37.
- Calarese T, Ferlazzo E, Daquin G, Genton P, Di Bella P, Villeneuve N. Ictal paresis associated to PLEDs in two children: a video-EEG study. *Seizure* 2008; 17: 735-9.
- Fukuda M, Nishida M, Juh sz C, et al. Short-latency median-nerve somatosensory-evoked potentials and induced gamma-oscillations in humans. *Brain* 2008; 131: 1793-805.
- Matsumoto R, Ikeda A, Hitomi T, et al. Ictal monoparesis associated with lesions in the primary somatosensory area. *Neurology* 2005; 65: 1476-8.
- Nii Y, Uematsu S, Lesser RP, Gordon B. Does the central sulcus divide motor and sensory functions? Cortical mapping of human hand areas as revealed by electrical stimulation through subdural grid electrodes. *Neurology* 1996; 46: 360-7.
- Pohlmann-Eden B, Hoch DB, Cochius JI, Chiappa KH. Periodic lateralized epileptiform discharges: a critical review. *J Clin Neurophysiol* 1996; 13: 519-30.
- Tseng YL, Chen YF, Lai SL. Ictal paralysis with tonic eye gazing mimicking a pontine infarction. *Seizure* 2006; 15: 637-42.
- Villani F, D'Amico D, Pincherle A, Tullo V, Chiapparini L, Bussone G. Prolonged focal negative motor seizures: a video-EEG study. *Epilepsia* 2006; 47: 1949-52.