Clinical commentary

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Unilateral abdominal clonic seizures of parietal lobe origin: EEG findings

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ABSTRACT – Unilateral abdominal clonic seizures represent a peculiar and rare manifestation of focal onset epilepsy. We present the case of a 26-year-old man with right-sided abdominal clonic movements associated with seizures arising from the left parietal area. We show the ictal EEG correlates of these events, including source localization of early ictal spikes; findings that have not been demonstrated in previously reported cases. The electro-clinical features in this patient suggested that clinical onset occurred after anterior propagation of ictal activity from a region posterior to the neck and trunk area of the sensory homunculus of the postcentral gyrus.
[*Published with video sequence on www.epilepticdisorders.com*].

Key words: abdomen, epilepsy, homunculus, source localization, trunk, clonic seizures

Unilateral abdominal or truncal seizures are rarely seen and have been described in only a small number of published reports. Previous reports of these seizures have not demonstrated definite EEG correlations except in some cases of epilepsia partialis continua (EPC), and localization of the epileptogenic area responsible for abdominal motor seizures has been controversial. A brainstem localization was initially postulated at the turn of the last century (Jackson and Singer, 1902; Jackson and Barnes, 1902). However, case studies over the past 50 years, based mainly on the presence and site of associated structural lesions, have reported hemispheric localizations in contralateral frontal (Rosenthal *et al*., 1986; Rosenbaum and Rowan, 1990; Oster *et al*., 2011), frontoparietal (Chalk *et al*., 1991; Fernández-Torre *et al*., 2004; Dafotakis *et al*., 2006), parietal (Matsuo, 1984; Tezer *et al*., 2008), parieto-occipital (Nehlil and Thurel, 1967), or even occipital (Ribeiro *et al*., 2015) regions.

Ictal EEG findings have largely been limited to the demonstration in EPC cases of periodic lateralized epileptiform discharges (PLEDs)

VIDEO ONLINE

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over contralateral frontoparietal (Rosenbaum and Rowan, 1990; Chalk *et al*., 1991; Fernández-Torre *et al*., 2004; Tezer *et al*., 2008) or occipital (Ribeiro *et al*., 2015) areas. The EPC patient of Ribeiro *et al*. (2015) with occipital PLEDs, however, was shown to have electrographic transitions to fast rhythmic occipital discharges lasting 20-60 seconds, which were accompanied by an increase in frequency of contralateral abdominal jerking. Occipital lobe seizures leading to abdominal motor movements presumably signify anterior propagation of the ictal activity. Ribeiro *et al*. (2015) also speculated that individual variability in homuncular organization and cortical neuroplasticity may have played a role in their EPC patients, both of whom presented years after haemorrhagic or ischaemic stroke. Ictal epileptiform changes have not been reported in non-EPC cases, although postictal EEG changes over the frontocentral region were described in one patient (Matsuo, 1984).

We present here the case of a patient with chronic medically refractory epilepsy and unilateral abdominal seizures with associated ictal EEG changes recorded over the contralateral parietal region. Source localization of interictal and early ictal spikes, combined with initial clinical symptoms and signs, suggested an epileptogenic region posterior to the neck and trunk area of the sensory homunculus of the postcentral gyrus.

Case study

The patient was a 26-year-old, right-handed man with a history of medically refractory seizures since the age of 16 years. He was born prematurely at 31 weeks from non-consanguineous parents, but development was normal.

Brain MRI at age 18 years demonstrated atrophy and gliosis affecting the left peri-insular region and surrounding parietal area with ex-vacuo dilatation of the Sylvian fissure. The MRI findings were unchanged on repeat scans at ages 20, 22 and 24 years.

His main seizures were characterized by clonic movements of the right side of the abdomen, preceded by an uncomfortable "pulling" sensation involving the right side of the neck and shoulder, with or without a few clonic movements of the right arm and head deviation toward the right side (*see video sequence*). These seizures occasionally progressed to secondarily generalized tonic-clonic convulsions. Non-generalized seizures typically lasted less than two minutes and were followed by postictal drowsiness and rarely postictal right-sided paresis.

A second, less frequent seizure type consisted of episodes of behavioural arrest and blank stare, followed by unresponsiveness and manual automatisms. There had been no significant change in recent years in seizure semiology or seizure frequency (1-4 events per month), despite multiple trials of antiepileptic medications at maximum tolerated therapeutic dosages. The family history was remarkable only for a paternal uncle with seizures. The patient's neurological examination was normal.

The patient was admitted to the epilepsy monitoring unit (EMU) for characterization and localization of his events, with a view to potential surgical management. His AED regimen on admission included levetiracetam 750 mg BID, oxcarbazepine 1,500 mg BID, and clobazam 20 mg BID. Under tapering dosages of AEDs, five of his typical events, involving right-sided abdominal wall clonic movements, were recorded during one week of monitoring.

In one event, the patient spontaneously described the progression of the event to the video camera, from the earliest sensations in the neck and "shoulder blade", prior to the onset of his lateralized abdominal contractions ("... and the pulling comes..."), which he indicates in the video by lifting his shirt and pointing to the affected area (*see video sequence*). Examination of the video recordings revealed associated clinical motor manifestations involving early tonic or low-amplitude clonic contractions of the right side of the head, neck, and shoulder region, and later jerky movements of the right arm. Two of the events evolved to secondarily generalized tonic-clonic seizures.

Electrographically, the ictal onsets were heralded by sequential sharp waves recorded over the left mid parietal area (maximal amplitude in common average referential montage at P3>Pz>C3, O1>Cz, T5), evolving into a rhythmic theta and later alpha frequency ictal pattern recorded over the left centroparietal (P3, Pz, C3) region (*figure 1*). The two major motor seizures showed further anterior propagation to the left frontal region (F3) prior to secondary generalization. One different type of clinical seizure was recorded during the same monitoring period, typical of his second seizure type, marked by a blank stare and upper extremity automatisms, with ictal onset localized to the left anterior temporal region (F7, T3, F9, and Sp1).

Interictal epileptiform discharges were characterized by spike and wave or sharp and slow wave discharges, recorded maximally over the left mid parietal region (phase reversal in longitudinal and transverse bipolar montages at P3), with a topographic field extending to the left occipital region greater than central and posterior temporal regions. These interictal discharges appeared topographically identical to the early ictal sharp waves. Less frequent independent sharp and slow wave discharges were recorded over the left

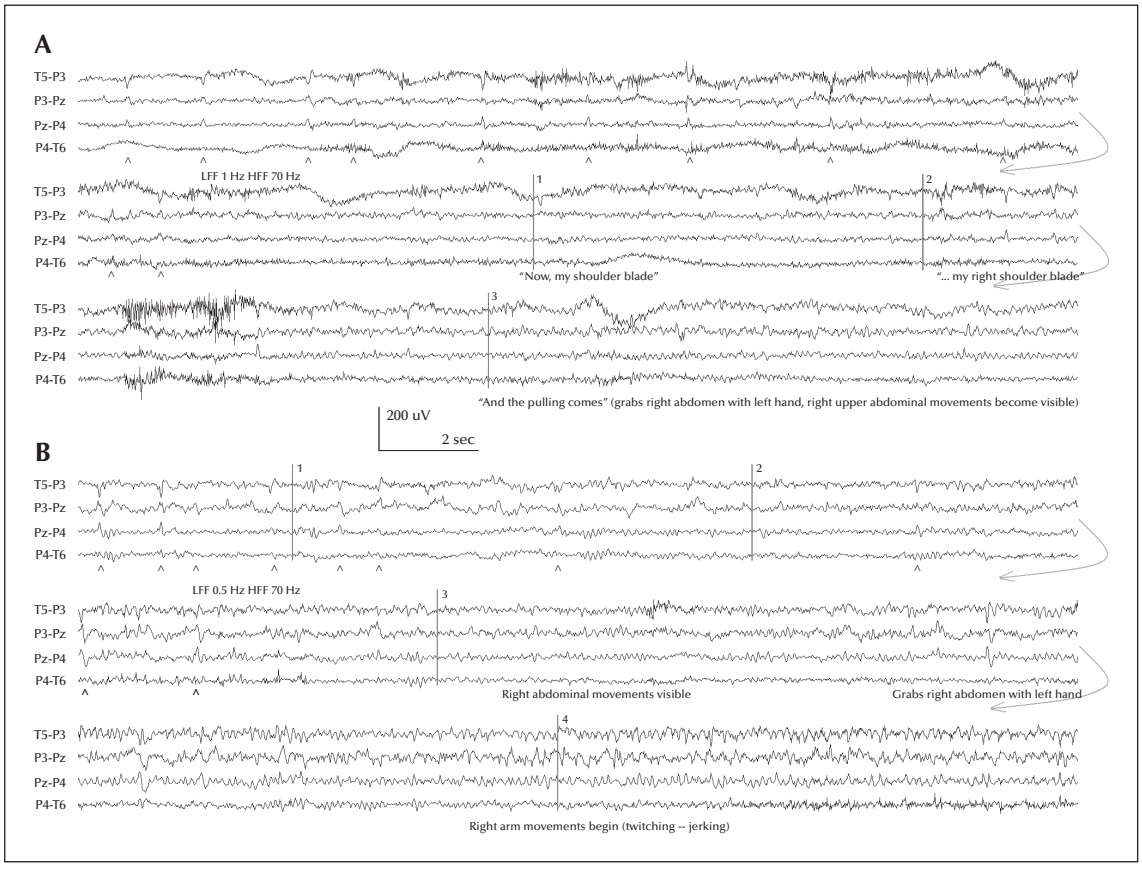

Figure 1. Ictal EEG findings shown in coronal bipolar chain across the parietal region.
Vertical lines mark the onset of the consecutive clinical features of the seizures: 1=shoulder/neck sensation; 2=neck/head/shoulder contractions; 3=abdominal contractions; and 4=arm contractions.
(A) Ictal onset heralded by sequential left mid-parietal sharp waves phase reversing at P3 (^), followed by the development of a rhythmic ictal theta-alpha pattern between P3-Pz, evident approximately five seconds before the abdominal movements begin (this is the seizure shown in the video sequence). Low-frequency filter (LFF): 1 Hz; high-frequency filter (HFF): 70 Hz.
(B) Ictal onset of another seizure showing again a run of sequential sharp waves over P3 (^), evolving into a rhythmic ictal theta-alpha pattern, evident around the time that abdominal movements first become visible (underneath the patient's shirt in this event). This seizure proceeded to secondary generalization. LFF: 0.5 Hz; HFF: 70 Hz.

anterior temporal region (phase reversal in longitudinal bipolar montage at F7 and F9 and at Sp1 in a circumferential transverse bipolar montage), the topographic field extending posteriorly to the left mid temporal region.

EEG-EMG polygraphic recordings were not obtained as the patient's EMU admission had been organized as a routine investigation for possible epilepsy surgery, precluding EEG-EMG back-averaging analysis of the abdominal clonic movements.

Recent brain MRI showed no change in the cortical atrophy involving the left parietal and peri-insular region (*figure 2A*). Functional MRI, motor, and somatosensory evoked potential studies (to precisely map motor and sensory function, in case of reorganization secondary to the cortical atrophy) were not performed given the routine nature of the clinical investigation and the patient's normal neurological examination.

Non-invasive source localization was performed on the early ictal sharp wave discharges using CURRY 6 (Compumedics, Abbotsford, Australia) and methods previously described (Wennberg and Cheyne, 2014). Thirty-four artefact-free sharp waves were averaged for modelling (*figure 2B*). Noise was estimated as the variance in the EEG signal over the interval from -550 to -125 ms before the peak of the averaged waveform. A 3-shell spherical forward model was used (a realistic volume conductor could not be created as the patient's MRI scan did not include acquisition slices through the top of the skull), and inverse modelling performed using a fixed coherent (equivalent current) dipole model. The dipole source solution was situated in the left parietal region, at least one gyrus

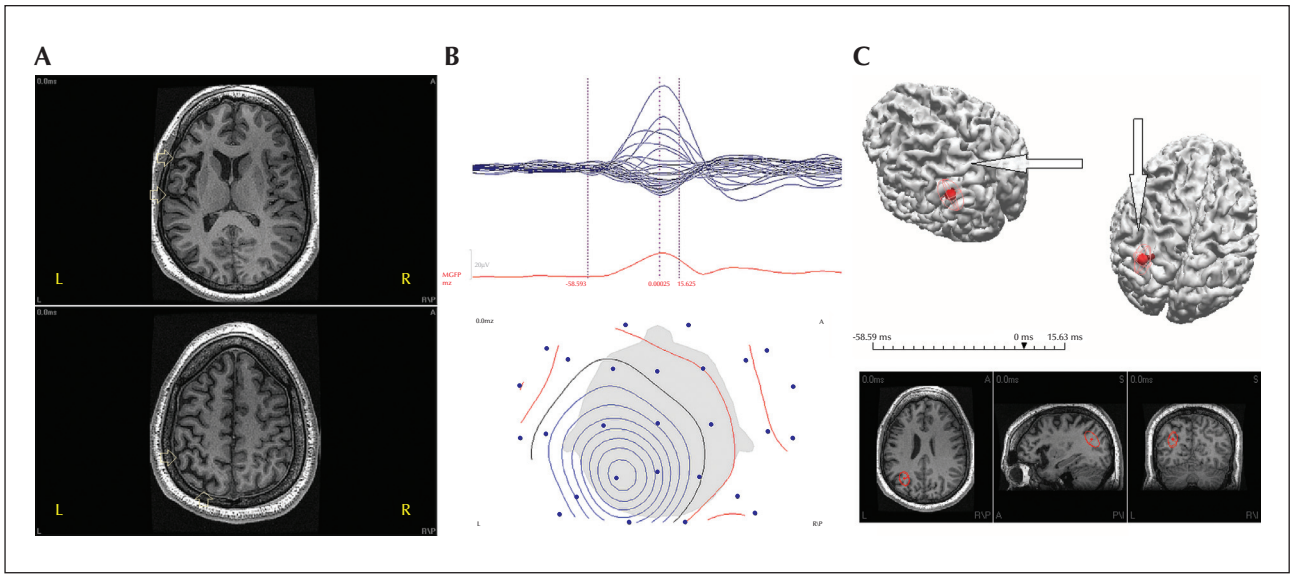

Figure 2. (A) Axial brain MRI demonstrating areas of left peri-insular and parietal atrophy (arrowheads). (B) EEG butterfly plot (upper image) and associated voltage topographic plot (lower image) of 34 averaged early ictal mid-parietal sharp waves (topographic plot at waveform peak, t=0). MGFP=mean global field power. (C) Dipole source solution of the averaged early ictal sharp waves, located in the region of the posterior wall of the parietal lobe gyrus posterior to the postcentral gyrus. Modelling performed on spike epoch extending from just before onset to just after peak (outer vertical lines in [B]). Volume conductor: 3-shell spherical model. LFF: 1 Hz; HFF: 30 Hz. Explained variance (goodness-of-fit): 97.7%. Arrows=postcentral gyrus.

posterior to the postcentral gyrus (*figure 2C*). A distributed source inverse model (sLORETA) returned a parieto-occipital source solution situated 15 mm posterior to the dipole source solution (*supplementary figure 1A*). Surface topographic voltage maps suggested anterior propagation of the sharp wave discharges over a period of 30 ms in a parieto-occipital greater than posterior temporal (O1, P3>T5) to mid parietal direction (P3>Pz) (*supplementary figure 1B*).
In light of the proximity of ictal EEG localization to primary sensorimotor cortical regions and the detection of an independent site of clinical seizure onset in the anterior temporal region, the patient has decided to forego further pre-surgical investigations at this time.

Discussion

The cortical representation of the trunk comprises a very small area compared to the arm, hand, and face regions of the motor and sensory homunculi (Penfield and Jasper, 1954). Along the motor homunculus, the trunk is situated between the shoulder and the hip. On the sensory homunculus, from lateral to medial, the sequence of cortical representation is: arm, shoulder, head, neck, trunk, hip, and leg (Penfield and Jasper, 1954). In our patient, initial clinical symptoms and signs affecting the right neck, head, and shoulder area, just prior to the onset of right-sided abdominal symptoms and signs, along with early ictal EEG changes recorded over the left parietal area, localized the site of seizure onsets posterior to the central sulcus. The clinical features of the seizures, and the associated EEG findings, indicated anterior and medial propagation of ictal activity first to the trunk area of the sensory homunculus and then, presumably, further anterior propagation to the trunk area of the motor homunculus.
Partial onset seizures that involve unilateral truncal muscles are rare. Epileptic abdominal movements have been described in the literature over the past half-century as myoclonic or clonic movements or motor spasms, mainly identified as part of status epilepticus, with most of the reported cases presenting as EPC, usually associated with one or more structural lesions in contralateral brain regions (Nehlil and Thurel, 1967; Rosenthal *et al.*, 1986; Matsuo, 1984; Rosenbaum and Rowan, 1990; Chalk *et al.*, 1991; Fernández-Torre *et al.*, 2004; Tezer *et al.*, 2008; Oster *et al.*, 2011; Ribeiro *et al.*, 2015). The locations of the lesions ranged from frontal to occipital areas, and the nature of the lesions was variable, including infectious causes (Matsuo, 1984; Rosenthal *et al.*, 1986; Chalk *et al.*, 1991), neoplasms (Matsuo, 1984; Rosenbaum and Rowan, 1990; Fernández-Torre *et al.*, 2004), cortical dysplasias (Tezer *et al.*, 2008; Oster *et al.*, 2011), and cerebrovascular

disease (Ribeiro *et al*., 2015). Another report described a case of abdominal status myoclonus in postanoxic coma; this was felt to be of subcortical origin and had no EEG correlation (Legriel *et al*., 2012).

Ictal EEG findings have, to date, been described only in cases presenting as EPC. Ictal epileptiform changes have not been reported in non-EPC cases; an observation attributed to either the small cortical representation of the trunk on the motor cortex or possibly to obscuration of the EEG by muscle and movement artefact (Matsuo, 1984; Oster *et al*., 2011; Ribeiro *et al*., 2015).

As described in the introduction, presumptive cortical localizations of past cases of abdominal seizures (EPC and non-EPC), based on structural, metabolic, and/or EEG findings, have included frontal (precentral gyrus), parietal, and even, perhaps surprisingly, occipital areas. To reconcile these different localizations, it is reasonable to consider that either direct ictal generation in the truncal area of the motor homunculus or activation of this motor area secondary to propagated ictal activity from a nearby region (*e.g*. somewhere near the truncal region of the sensory homunculus in the parietal lobe) could produce the same end effects of abdominal movements. It must also be acknowledged that motor contractions have long been known to be elicitable by direct stimulation of the postcentral gyrus (Penfield and Jasper, 1954).

In our patient, it seems clear that ictal onsets occurred in the parietal lobe. Nonetheless, we do not mean to suggest that all cases of abdominal seizures will be localizable to postcentral epileptogenic areas. Anterior propagation of ictal activity from parietal or parieto-occipital areas, as well as posterior propagation from frontal areas, if perfectly situated, could equally produce the clinical features of abdominal motor seizures.

The rarity of the presentation as a partial seizure manifestation presumably reflects a requirement for a very focal ictal discharge to arise within, or propagate to involve precisely the very small truncal areas of the homunculi. The minuteness of the surface representation of the homuncular trunk areas likely explains the absence of ictal EEG findings in previously reported non-EPC cases of abdominal motor seizures. In our patient with anterior propagation from a parietal epileptogenic area, ictal onsets initially involving a larger area of association cortex facilitated the recording of EEG changes preceding and during propagation toward the truncal regions of the homunculi. □

Supplementary data.

Summary didactic slides and supplementary figure are available on the www.epilepticdisorders.com website.

Disclosures.

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

Legend for video sequence

One of the patient's typical left parietal onset seizures involving right abdominal motor movements. The patient can be heard describing the progression of the clinical episode, which initially involved an abnormal sensation and some brief tonic or clonic contractions affecting the right neck and shoulder area, prior to the onset of his right-sided abdominal sensation ("... and the pulling comes ..."); and then the visible abdominal movements.

Key words for video research on www.epilepticdisorders.com.

Phenomenology: focal seizure not otherwise specified
Localization: posterior cortex (parietal), sensorimotor cortex
Syndrome: focal non-idiopathic parietal
Aetiology: brain malformation (not specified)

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TEST YOURSELF

(1) Along the motor homunculus, the trunk is situated between which two areas?

(2) Along the sensory homunculus, what is the sequence of the following six areas:
neck, arm, leg, trunk, head, shoulder?

(3) Abdominal seizures may have ictal onset in which cortical areas?

Note: Reading the manuscript provides an answer to all questions. Correct answers may be accessed on the website, www.epilepticdisorders.com, under the section "The EpiCentre".