Transient parietal hand syndrome after cortical resection

Joel M. Oster 1, Diana Apetauerova 2, Christine Thomas 3, G. Rees Cosgrove 4

1 Department of Neurology, Lahey Clinic Medical Center, Burlington; Tufts University, School of Medicine, Boston, MA
2 Department of Neurology, Lahey Clinic, Burlington, MA
3 Department of Pathology, Lahey Clinic, Burlington, MA
4 Department of Neurosurgery, Warren Alpert School of Medicine, Brown University, Providence RI, USA

Received February 8, 2011; Accepted July 5, 2011

ABSTRACT – This case study with video recording describes a brief neurological examination of a rare post-surgical finding in a patient with intractable seizures who had a right parietal topectomy in order to cure focal and disabling epilepsy. Contemporaneously during the resection and while awake intraoperatively, the patient developed features characterised by involuntary, purposeless, and almost ballistic movements of the contralateral left upper extremity. These involved the shoulder and, more distally, the arm with less involvement of the hand itself and some clonic movement at the elbow, persisting for approximately 24 hours after surgery. Although identified in only one case in our series, we have named the resulting clinical phenomenology “parietal hand syndrome”. A Medline search does not reveal any other such case in the English literature with the clinical elements and actual video documentation of neurological examination noted in our case report in the immediate post-operative setting. In this regard, this is a unique clinical report. [Published with video sequences]

Key words: epilepsy surgery, refractory seizures, parietal resection, parietal hand, parietal dysfunction

Case study

A 16-year-old right-handed patient with refractory epilepsy of four years and normal neurodevelopmental history was referred for surgical treatment of epilepsy having been unsuccessfully treated with multiple antiepileptic medications.

Preoperatively, the patient underwent a video-EEG study which identified brief clinical events in which she had an abnormal sensation, lasting for approximately 10 seconds, in the palm of her left hand and, if prolonged, could progress to tonic and clonic contractions of the left hand and arm. These events correlated with only subtle, if any, EEG changes seen maximally in the right centro-parietal derivations (figure 1).

Preoperative MRI was reported to be negative by other centres with no definite structural abnormality, although we identified that there was a notable area of gyral thickening in the right parietal cortex and
we speculated that this might be a cortical dysplasia causing the seizures (see figure 4). Multiple peri-operative ictal spectroscopy studies and interictal 18-FDG PET were either negative or non-specific for localisation. Subsequent invasive video-EEG monitoring verified the focal ictal onset electrical disturbance in contacts over the superior right parietal lobe immediately posterior to the somatosensory strip representation of the hand (figure 2). The patient subsequently underwent a focal parietal resection under local anaesthesia immediately posterior to the somatosensory strip in the superior parietal region and almost instantly developed involuntary, purposeless, and uncontrollable myoclonic movements of the left arm and hand which persisted for approximately 24 hours, as noted above. The patient seemed unaware of these movements. This was accompanied by sensory ataxia upon reaching out.

Post-surgically, the patient appeared to have only mild cortical sensory loss in the left hand at the approximately six-month post-operative visit. There was otherwise no weakness or upper motor neuron signs noted during this time period. The abnormal movements were not associated with any changes on electrocorticography performed contemporaneously with the resection and the abnormal movements. The video examination that was subsequently recorded in the recovery suite the following morning illustrates the transient movement disorder of the left upper extremity, although substantially less dramatic than seen during surgery and the immediate post-operative period (see video sequence). Pathology identified large atypical neurons and neuronal processes with abnormal laminar organisation consistent with focal cortical dysplasia (figure 3). Pre- and post-operative MRI illustrated the extent of resection (figure 4). With more than five years of follow-up, the seizures have not recurred.

**Figure 1.** Surface EEG; onset of a typical seizure. Note there are only subtle changes at onset of the seizure (see arrows). After approximately the third second, there is semi-rhythmic irregular theta most prominent in the right centro-parietal derivations. After about the sixth second, there are possible spikes seen in the same regions.

**Figure 2.** Invasive EEG monitoring identifying focal onset of seizure activity in the right parietal convexity at one derivation (see arrow).

**Figure 3.** Cross-section of resected specimen stained with haematoxylin and eosin; note the pathology is consistent with cortical dysplasia.
Transient parietal hand syndrome

Discussion

The exact physiology observed in this case is unclear but a review of the literature did identify some theoretical reasons for the unusual movement disorder observed in this case. The clinical manifestations of parietal resections in patients with intractable epilepsy have been reviewed and described in the literature (Salanova et al., 1995; Ajmone-Marsan and Goldhammer, 1973; Cascino et al., 1993). Lesions of the right parietal lobe cause varied somatosensory impairments and distinct motor disruption and can include hemi-neglect, impairment of visuo-spatial tasks, and difficulty in performing complex motor tasks, including apraxia and ataxia (Salanova et al., 1995; Ajmone-Marsan and Goldhammer, 1973; Cascino et al., 1993; Mesulam, 1981). Patients with right parietal lesions may be unaware of their impairments and may have perceptual agnosias in which the patient is unable to perceive certain patterns of perceptual loss (Salanova et al., 1995; Ajmone-Marsan and Goldhammer, 1973; Cascino et al., 1993; Mesulam, 1981; Hecaen et al., 1956; Critchley, 1953). Some authors postulate that parietal lesions interfere with motor planning and control and may secondarily cause dys- function (Gordon, 1999; Jeannerod et al., 1984, 1995; Binkofski et al., 1998, 2001; Jancke et al., 2001, Wolpert et al., 1998; Weiss et al., 2001; Freund, 1987, 2003; Knecht et al., 1996). There may be activation of specific parietal regions with task-specific activities (Binkofski et al., 1998; Freund, 2003; Knecht et al., 1996). Activation of neurons may occur in the parietal lobe prior to neuron pools in the motor cortex indicating that there is some “pre-motor” or “planning” process that takes place prior to movement. It has been suggested that there are neurons that may be activated even in response to contemplation of a movement and that the “readiness potential” or Bereitschafts potential might be a marker of this phenomenon (Jahanshahi and Hallet, 2003). In parietal syndromes, two-point discrimination may be impaired and astereognosis may exist (Freund, 1987, 2003; Knecht et al., 1996). If the lesion is in the right parietal lobe, bilateral impairments in graphesthesia occur (Knecht, 1987; Freund, 2003; Jahanshahi and Hallet, 2003). In more anterior parietal lesions, two-point discrimination impairment correlates with absence of N20 peak on somatosensory evoked potentials (Freund, 2003; Knecht et al., 1996; Jahanshahi and Hallet, 2003). Reports indicate that relatively severe motor impairments are noted particularly with left parietal lesions (Weiss et al., 2001). Based on the above literature, it might be inferred that the motor difficulties seen with parietal lesions may be due to an impaired internal spatial-temporal body image caused by deafferentation or by damage to tracts that involve modulation of efferent motor systems. While our patient had no pre-existing parietal dysfunction prior to the focal parietal resection, we speculate that the transient motor manifestations seen in our case may be related to another syndrome termed the “Alien Hand Syndrome” (AHS), first identified in 1908 (Legon and Staines, 2006; Scepkowski and Cronin-Golomb, 2003; Banks et al., 1989). Patients with the AHS syndrome possess a hand that exhibits movements that are not under voluntary control of the patient. AHS is a chronic and progressive condition in contrast to the transient phenomenology exhibited by our patient, although some cases occur abruptly in anterior cerebral artery stroke syndromes. The movements in AHS are described as grasping movements in the non-dominant hand. The alien hand may also engage in purposeful-type movements, such as using tools, or manipulating clothes. According to the literature, the pathologic hand of AHS may be able to exert excellent muscle power, although patients cannot

Figure 4. Panel 1: preoperative T1-weighted SPGR coronal sequence MRI with imaging through area of interest. The depth of a noted sulcus appears thickened in this region (arrow). Panels 2 and 3 illustrate T1 axial and coronal MRI imaging with and without gadolinium contrast, respectively. Arrow and caliper markings indicate the resected region in panels 2 and 3 corresponding to ictal onsets in the superior parietal lobe immediately posterior to the somatosensory strip.
effectively direct their voluntary actions (Scepkowski and Cronin-Golomb, 2003; Banks et al., 1989; Feinberg et al., 1992; Victor and Ropper, 2001). Attempts to move a limb in order to accomplish a purposeful act might result in a totally inappropriate movement. Alternatively, the limb may “drift off” and assume an odd posture such that patients may sense the arm is beyond their control although they recognize the extremity as their own. While there is no treatment for AHS, the literature indicates that symptoms may be relieved by giving the hand an object to manipulate and thereby keep it occupied (Scepkowski and Cronin-Golomb, 2003; Banks et al., 1989; Feinberg et al., 1992; Victor and Ropper, 2001).

Alien Hand Syndrome cases exhibit parietal lobe pathology, but in contrast to our case, this is not exclusive (Scepkowski and Cronin-Golomb, 2003; Banks et al., 1989; Feinberg et al., 1992, Victor and Ropper, 2001). AHS has been described as a side effect of resection in various regions that may have parietal fibre connections in the white matter, such as in the anterior corpus callosum. Posterior cerebral lesions without lesions of the corpus callosum have also been noted. Patients with extra-pyramidal or neurodegenerative diseases such as cortical-basal ganglionic degeneration (CBGD) may also present with AHS (Scepkowski and Cronin-Golomb, 2003; Banks et al., 1989; Feinberg et al., 1992; Victor and Ropper, 2001). Victor and Ropper identified, by postmortem examination, that several of the cases of CBGD had cortical atrophy mainly in the frontal motor, premotor, but notably the anterior parietal lobes (Victor and Ropper, 2001). AHS in this patient’s clinical and postmortem findings, referred to as PHS in this report, have significant parietal lesions or pathology, we postulate that perhaps these conditions exhibit different clinical phenomenology, depending on which additional brain structures are involved. Since parietal lobe function is important in sensing body image, location, and position or posture, and the parietal lobe and its connections are involved in the planning and modulation of movement, as noted in the cited literature, we speculate that the surgical resection performed in our case at the superior parietal region immediately posterior to the somatosensory cortex, representing the hand and upper extremity, resulted in either functional deafferentation of the sensori-motor system or disruption of the efferent modulation of the motor system.

We are uncertain as to why the PHS phenomenology was self limited to a brief, approximately 24-hour period. Perhaps the ballistic qualities were manifested by the acute deafferentation of the subthalamic nucleus from parietal cortical inputs and over the noted time course, the system adapted. Nonetheless, in our opinion, the immediate occurrence of these gross abnormal movements upon resection of the superior parietal cortex implicates this area as the primary cause of the parietal hand syndrome in humans, as noted in this case.

Disclosure.
None of the authors has any conflict of interest or financial support to disclose.

Legend for video sequence

Post-operative examination of the patient with parietal hand syndrome.

Key words for video research on www.epilepticsendnotes.com

type I

Phenomenology: AURA (autonomic), AURA (somatomotor), tonic posture

Localization: posterior cortex (right)

Syndrome: focal non-idiopathic parietal

References


Transient parietal hand syndrome


