Anatomo-electro-clinical correlations: the University of Cleveland, USA Case Report
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Medically intractable seizures originating from the primary somatosensory hand area

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ABSTRACT – [Case records of Epileptic Disorders. Anatomo-electro-clinical correlations. Case 03-2008] A 33-year-old woman had begun having intractable somatosensory seizures affecting the left hand since the age of 13 years. Occasionally, her seizures progressed to left arm posturing followed by secondary generalization. Scalp EEG revealed interictal epileptiform discharges in the right posterior quadrant, but with no ictal EEG correlates. Brain MRI showed a right temporal encephalomalacia, sparing mesial temporal structures, suggestive of a perinatal vascular insult. Ictal electrocorticogram, electrical stimulation mapping, and somatosensory evoked potentials localized the ictal onset to the hand area of the postcentral gyrus. Resection of that area resulted in total resolution of seizures with no significant lasting deficits. Potential complications of resecting the primary somatosensory hand area can be severe, as proprioceptive sensory loss may be permanent, resulting in significant disability. Such deficits may be temporary however, and the literature continues to report conflicting results regarding postsurgical outcome. Cortical plasticity may explain recovery of sensory deficits after partial resection of the primary somatosensory hand area. Multiple subpial transections of that area are sometimes performed to minimize functional deficits, but seizure control may be less optimal than with cortical resection.

Key words: somatosensory seizures, intractable epilepsy, cortical mapping, epilepsy surgery, cortical plasticity

Pure somatosensory seizures may originate from the primary and secondary somatosensory cortices, and supplementary motor areas (Penfield and Jasper 1954), with neurological differences depending on the area of origin. They are less likely to be associated with ictal changes on scalp EEG than seizures accompanied by motor manifestations (Devinsky et al. 1988), possibly because motor seizures reflect more expansive propagation of the ictal discharge. Thus, pure somatosensory seizures frequently necessitate invasive monitoring for localization (Devinsky et al. 1989). Unlike the face, which is bilaterally represented in the primary somatosensory cortex (Lehman et al. 1994), the unilateral representation of the hand in the contralateral postcentral gyrus makes resection particularly hazardous as it may result in sensory apraxia and proprioceptive sensory loss.

The outcome after resection of the primary somatosensory hand area has varied between favorable and detrimental. Penfield and Erickson believed that resection of this area could be as problematic as resecting the motor arm area, as it risks rendering the hand dysfunctional for delicate tasks (Penfield and Erickson 1941). Other authors also reported permanent deficits resulting from resection of the
primary somatosensory arm area (Cohen-Gadol et al. 2003, Pilcher et al. 1947). However, there are reports that astereognosis and proprioceptive sensory loss associated with resection of the postcentral gyrus can be reversible (Pilcher et al. 1947); a number of authors have mentioned favorable outcome after resection of primary somatosensory cortex for treatment of neoplasms (Gregorie and Goldring 1984), seizures in the setting of a dysembryoblastic neuroepithelial tumor (DNET) (Asano et al. 1999), and nonlesional epilepsy (Cohen-Gadol et al. 2003), as well as in unselected cohorts with perirolandic surgery (Pondal-Sordo et al. 2006). Multiple subpial transections (MST) in patients with intractable focal epilepsy originating from eloquent cortex emerged as an alternative technique that aimed at minimizing functional deficits, but it has been associated with less favorable outcome than cortical resection (Spencer et al. 2002). Additionally, MST may be associated with a higher chance of late seizure recurrence than resective surgery (Orbach et al. 2001). There are no clearly defined predictors to help decide whether resection of the primary somatosensory hand area will result in lasting deficits. However, lesions occurring earlier in life may correlate with better outcome after resection in view of the higher degree of cortical plasticity (Graveline et al. 1998). Additionally, it has been reported that multiple representations of the hand area in the primary somatosensory cortex may be the basis of the favorable outcome after limited cortical resections (Asano et al. 1999, Cohen-Gadol et al. 2003, Gregorie and Goldring 1984). Thus, it is very likely that complete resection of the somatosensory hand area will result in a profound and permanent loss of hand cortical sensation, whereas partial resection will only produce partial deficits with a good potential for recovery. This cannot be clearly discerned from the literature since most authors do not report a detailed account of the extent and depth of the resection of the somatosensory area. Seizure freedom without lasting sensory deficits after resection of the primary somatosensory hand area have only been reported rarely (Asano et al. 1999, Gregorie and Goldring 1984). We add to the literature our experience of a patient with seizures originating from the primary somatosensory hand area, in whom corticectomy of that area resulted in elimination of her seizures, without significant lasting deficits.

Case presentation

History

The patient is a 33-year-old, right-handed woman with normal developmental milestones and no perinatal complications. Her seizures started at the age of 13 years, and had been refractory to medical treatment. Most seizures consisted of left hand somatosensory symptoms, which she described as “pulsing”, “tightening” or “curling” lasting 10 to 30 seconds, without alteration of awareness. This sensation affected mainly the left hand, but occasionally spread to involve the left arm. At times, these sensations were followed by left arm posturing, left head deviation, and secondary generalized clonic seizures. These seizures occurred almost daily despite therapy with various combinations of anticonvulsants. At the time of evaluation, she was receiving oxcarbazepine and lamotrigine. Previous anticonvulsants used have included levetiracetam, felbamate, and carbamazepine, all of which she discontinued because of suboptimal seizure control. The patient’s general and neurological examinations were normal.

Presurgical workup

Video-EEG monitoring with scalp electrodes demonstrated intermittent slowing in the right posterior quadrant, maximum over the P8 electrode location. Abundant sharp waves were seen in the same distribution (figure 1). The patient experienced 14 habitual seizures during monitoring, all manifesting as somatosensory experience in the left hand, with one seizure evolving into left arm tonic posturing followed by a secondary generalized clonic seizure. The ictal EEG during her somatosensory auras showed no discernible deviations from baseline.

Figure 1. Bipolar EEG montage using the standard 10-20 system showing sharp waves (arrows) in the right posterior head regions.
Quad-coil high-resolution brain MRI was obtained in a fashion described in a previous report from our epilepsy center (Goyal et al. 2004). The study showed a large focus of encephalomalacia in the right temporal lobe, sparing the mesial temporal structures (figure 2). However, loss of internal architecture in the right hippocampus and volume loss of the right hippocampal head and fornix were noted. Although the patient reported normal birth and develop-

Figure 2. Sagittal T1-weighted and horizontal FLAIR MRI sequences demonstrating a large focus of encephalomalacia in the distribution of the right middle cerebral artery sparing the right hippocampus and most of the right amygdala.
ment history, these findings were thought to be related to a perinatal vascular insult involving the distribution of the right middle cerebral artery.

Neuropsychological evaluation was performed with the results of all tests being accorded a mean standard score of 100, with a standard deviation of 15. Her full scale IQ was 88, verbal IQ 106, and performance IQ 91. She scored 97 on immediate verbal recall, 80 on delayed verbal recall, 97 on immediate visual recall, and 97 on delayed visual recall. The patient did not undergo an intracarotid amobarbital procedure, but underwent a functional MRI (fMRI) study for identification of language and motor regions. For the fMRI, sequential echoplanar BOLD images were obtained during alternate activation and rest periods. Paradigms employed included sequential finger tapping, silent word generation and passive receptive listening. Functional data were then superimposed on anatomical images for review. The study lateralized language to the left hemisphere, with the word generation task resulting in activation in the left frontal operculum and left middle frontal gyrus, and the word receptive phase activating the left superior temporal gyrus and a small area in the left frontal lobe at the junction of the middle and inferior frontal gyri just superior and anterior to the left frontal operculum. During the motor task, there was activation of bilateral, precentral and postcentral gyri, the supplementary motor region, and bilateral superior cerebellar hemispheres.

The patient was discussed in the multidisciplinary epilepsy surgery conference at our center before surgery was decided upon. Two possible locations of the seizure focus were considered: the hand area of the primary somatosensory cortex, and a clinically silent, parieto-occipital area with possible anterior propagation of the ictal discharge to the perirolandic hand area. The latter possibility was considered in light of the predominance of interictal epileptiform discharges over the right posterior quadrant and their absence centrally. The possibility of a seizure focus in the secondary somatosensory area was considered less likely because of the localized nature of the somatosensory aura to the left hand. It was decided to implant a subdural grid of electrodes over the right central and parietal areas, to allow localization of the irritative and seizure-onset zones, and for mapping of sensory and motor functions.

During the surgery, the encephalomalacia was noted in the perisylvian region of the right frontotemporal cortex. Much of the lateral temporal lobe was missing, and a region of sclerotic and firm, brain parenchyma was observed in the parietal operculum just superior to this cavity. An 8 x 8 grid of platinum-iridium electrodes with an interelectrode distance of 1 cm was implanted over the right hemisphere as shown in figure 3.

Intracranial monitoring and surgery

Subdural electrode monitoring showed frequent epileptiform discharges from the right parieto-occipital region (figure 4A). These discharges consisted of biphasic or triphasic apiculate waveforms, with superimposed fast activity (100-125 Hz). Intermittent slowing was seen in the same distribution. No interictal epileptiform discharges were noted over the perirolandic area. Eight seizures were recorded, all consisting of left hand somatosensory aura,
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and one progressing to versive head deviation to the left, followed by tonic posturing and clonic movements of the left arm. The electrocorticographic ictal discharge was identical in all seizures, consisting initially of a fast frequency activity (80-120 Hz) at electrode #30, found later to overlie the primary somatosensory area of the hand, before evolving, during some seizures, into repetitive spikes also localized selectively over electrode #30 (figure 4B). Careful review of the video-EEG study found one-to-one correspondence between such gamma activity over electrode #30 and the patient’s report of the somatosensory aura, i.e. the fast activity was seen in association with every report of an aura, but not interictally.

Figure 4 depicts the locations of ictal and interictal discharges on the surface of the brain, as well as the results of electrocortical stimulation mapping (ESM). Somatosensory and motor areas of the left arm and face were found in the perirolandic area, and more posterior electrodes appeared to overlie visual association cortex as suggested by the visual phenomena elicited in that region. Of note, electrical stimulation of electrode #30 resulted in a left fourth and fifth finger tingling sensation at 6 mA that was not the same as the patient’s habitual seizures. Somatosensory evoked potentials stimulating the left median nerve showed phase reversals between electrodes 23 and 24, 31 and 32, 39 and 40, confirming the location of the central sulcus between the anterior two rows of the grid (figure 6). These findings were discussed again in the multidisciplinary epilepsy surgery conference, and two options were considered. Proponents of multiple subpial transections (MST) of the hand area argued that corticectomy of the area may result in debilitating proprioceptive sensory loss, whereas proponents of corticectomy argued that MST was less likely to result in optimal seizure control. These considerations were discussed with the patient who was adamant that she wished this region removed for optimal seizure control and was willing to accept permanent disability in the non-dominant hand if this optimized her chances of seizure control.

The patient was taken to the operating room where the electrode grids were removed, and numbered tickets were placed over each of the relevant numbers of the electrode grid. She was then fully awakened in the operating theater, and intraoperative electrocortical stimulation mapping confirmed the location of the primary motor and sensory strips. After removal of brain tissue underlying electrode #30, the patient reported numbness over the dorsum of the left hand and wrist, but continued to be able to use the left arm in a functional manner. Sensations over her palm appeared intact. In addition to the hand area of the post-central gyrus (electrode #30), regions of the cortex immediately posterior to the primary sensory cortex and along the encephalomalacic cyst were removed, as they were noted to be extremely gliotic, hemosiderin-laden, rubbery and tough (figure 7).
Post-operative outcome

On the first post-operative day, the patient showed extinction in the left hand to double simultaneous stimulation, and had difficulty locating her hand in space without visual guidance. Pinprick and temperature sensations appeared decreased in the left hand. These symptoms improved markedly within a week. One month after the surgery, she complained of distal left arm paresthesia, but examination revealed normal perception of vibration and light touch. Her pinprick sensation and joint position sense continued to be decreased in the left hand, and she clearly had impairment in two-point discrimination and agraphesthesia. She also had a dense, left, homonymous hemianopsia, but no motor deficits. Of note, the post-operative MRI showed a focus of diffusion restriction in the surgical bed in the right parietal lobe with corresponding low ADC signal, consistent with an area of small paren-
chymal infarct. This was felt to be the likely explanation of the patient’s hemianopsia, in view of the fact that areas along the posterior border of the encephalomalacic cyst that were resected were grossly gliotic and included only two of the eight electrodes were visual phenomena were elicited by ESM (#44, where the patient experienced a left hemifield “flicker” at 20 mA, and #51, where she saw “rainbow colors” in the left hemifield at 16 mA). Six months after surgery, her left homonymous hemianopsia was not changed, but her paresthesia had resolved and her perception of vibration, temperature and light touch were normal in the left hand. The deficits in pinprick perception

Figure 7. Post-operative coronal and horizontal T2 MRI sequences showing evidence of resection of right parietal and temporal areas. Extra-axial fluid and air are seen consistent with postsurgical changes. The mesial temporal structures were not included in the resection.

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became limited to a small area in the ulnar distribution of the left hand, and she had normal, two-point discrimination and no agraphesthesia. Her performance IQ dropped to 81. The patient continues to be seizure-free 10 months after the surgery.

**Discussion**

We present a case of medically intractable seizures originating from the hand area of the primary somatosensory cortex in whom awake craniotomy with limited surgical excision produced optimal seizure control without lasting deficits. The literature appears to contain conflicting results as regards the outcome after primary somatosensory cortical resection, with some authors reporting favorable outcome and others persistent deficits.

Cortical stimulation and resection of the perirolandic cortex were pioneered by Victor Horsley in the 1880s, who investigated the focal nature of convulsions and cortical localization of motor function. He produced accurate motor maps and performed a series of neocortical lesionectomies in patients with epilepsy (Edie 2005, Horsley 1886, Horsley 1890). J. Hughlings Jackson, who described focal motor seizures, worked closely with Horsley, and also entertained surgical excision as an option to rid patients of “worse than useless” cells in the primary motor area that resulted in frequent seizures (Jackson 1890). In 1896, Gowers agreed with Horsley that resection of cortical areas, whose stimulation produced initial ictal symptoms, would abolish seizures (Pilcher et al. 1947).

Horsley used electrical stimulation in anesthetized patients, so he mapped motor, but not sensory function. Indeed, towards the end of the 19th century, no clear cortical delineation of sensory and motor functions had been discerned, and some authors held that the rolandic cortex function was purely motor, while others believed it to be purely sensory (Penfield and Boldrey 1937). Grünbaum and Sherrington were the first to demonstrate the localization of the primary motor cortex to the prerolandic gyrus using unipolar faradization in 10 adult apes (Grünbaum and Sherrington 1901). They reported that movement ended abruptly behind the central sulcus, and commented on the somatotopic organization of the motor strip, with the leg area being medial and the arm lateral. Extirpation of the hand area in these animals resulted in severe weakness initially, but within few weeks they reported remarkable improvement of motor function, with the animals regaining their ability to use their hand to climb. These authors also noted that resection of the postcentral gyrus did not result in any weakness, but did not comment on its function.

Not only did Harvey Cushing map the motor cortex in more than 50 anesthetized patients, but, in 1908, he was the first to stimulate the postcentral gyrus in awake subjects (Cushing 1909). He commented: “if it will be possible in the future to pick out with an electrode, areas of the brain from which a sensory aura of a focal convulsion has originated, we shall have advanced a long way toward the possible operative localization of subcortical irritative lesions of the immediately postcentral field”. One of Cushing’s patients began having seizures at the age of 13 years, manifesting as right hand sensation followed by motor phenomena in the right hand and face. Cushing suspected a lesion in the left postcentral territory, and stimulated the postcentral gyrus during wakefulness, reproducing the patient’s aura. He made a large, exploratory incision in the postcentral gyrus looking for a lesion, but found no gross abnormalities. He reported that postoperative deficits related to the exploratory incision included sensory disturbances in the hand and forearm, with incoordination that worsened upon eye closure. These deficits resolved in a few weeks.

Since then, several authors have commented on the resection of the motor cortex, (e.g. Sachs 1935), with much less frequent reports about outcome after resection of the primary somatosensory cortex. In 1942, Penfield and Erickson wrote: “the disability resulting from removal of motor arm area is so great that we have rarely touched the precentral gyrus, and interference with the post-central gyrus is almost equally troublesome, because the hand becomes awkward and useless for delicate tasks” (Penfield and Erickson 1941). Penfield warned again of the risks of resection of the perirolandic arm area (Penfield and Rasmussen 1950), but reported that the contralateral tactile and two-point discrimination deficits resulting from removal of the face area in the postcentral gyrus may be marked initially, but resolve without sequela. 

Pilcher et al. (1947) reported surgical outcome after resection of motor areas in 41 patients with non-lesional focal motor seizures. Six of these patients underwent resection of parts of the postcentral gyrus because their seizure semiology included sensory phenomena. In all six patients, arm paresthesia lasted only 3-10 days postoperatively. In two patients, astereognosis and loss of position sense lasted only 7-10 days. Partial loss of sensations to tactile and painful stimuli disappeared after several months in three patients, but appeared to persist in the three other patients.

More recently, SSEPs and cortical stimulation were used, under general anesthesia, to localize the perirolandic area in 31 patients (Gregorie and Goldring 1984). The authors reported favorable outcome, with one patient experiencing no deficits after resection of the hand area in the postcentral gyrus. Another series reported complete recovery from sensory apraxia and cortical sensory deficits in three of four patients who underwent resection involving the postcentral gyrus (Cohen-Gadol et al. 2003). The postcentral gyral resection in that series did not involve the hand area in any of the four patients, but included the shoulder area in two patients and face area in the other two. The same authors later reported minor sensory deficits after resection of the leg area in the primary sensory cortex.
cortex (Cohen-Gadol et al. 2004). Finally, Pondal-Sordo et al. (2006) reported an unselected sample of 52 patients who underwent perirolandic surgery, including 20 patients with surgery involving the post-central gyrus. Novel neurological deficits occurred in half of their patients, most of which involved speech and motor function. Only one patient had pure sensory deficits, which were mild in nature. These authors also commented that corticectomy resulted in better seizure outcome than MST. We considered MST in our patient in order to decrease the risk of disabling sensory deficits. This procedure is performed using finer parallel incisions of the cortex, 5 mm apart, theoretically interrupting horizontal fibers that are needed for neuronal recruitment, thus preventing seizures without affecting vertical projections that are indispensable for eloquent function (Morrell et al. 1989). This approach may be more effective if combined with surgical resection of non-eloquent cortex than if performed alone (Hu�nagel et al. 2006). A better analysis of MST performed in six centers on 211 patients with medically intractable epilepsy, of whom 53 underwent MST without resection, found excellent outcome (>95% seizure reduction) in 68-87% of patients who had both procedures performed (Spencer et al. 2002). On the other hand, 62-71% of those who underwent MST without resection had a similar, excellent outcome. In that meta-analysis, neither age of seizure-onset nor location of MST were found to be significant predictors of outcome.

On the other hand, a later report found a lower chance of good seizure outcome after MST compared with surgical resections (Pondal-Sordo et al. 2006), and some authors found an increased rate of seizure recurrence after MST, despite initial favorable results (Orbach et al. 2001). Additionally, some authors suggested that gliosis and cortical injury resulting from MST may themselves be epileptogenic (Cohen-Gadol et al. 1997, Rouguet et al. 1996). A meta-analysis of MST performed in six centers on 211 patients with medically intractable epilepsy, of whom 53 underwent MST without resection, found excellent outcome (>95% seizure reduction) in 68-87% of patients who had both procedures performed (Spencer et al. 2002). On the other hand, 62-71% of those who underwent MST without resection had a similar, excellent outcome. In that meta-analysis, neither age of seizure-onset nor location of MST were found to be significant predictors of outcome.

While resection of the ictal onset zone is a known predictor of favorable outcome (Babb et al. 1974, Gloor 1975), no generalizations can be made about the value of resecting the irritative zone. Some authors believe that irritative zones need not be resected in order to achieve seizure control (Hu�nagel et al. 2000), and others have shown that such resections are associated with good surgical outcome (Armon et al. 1996). The reason we resected the irritative zone in our patient is twofold. Firstly, the patient’s irritative zone was close to a structural lesion and appeared grossly gliotic. Secondly, her epileptiform discharges consisted of spikes with superimposed ripples. Whereas interictal spikes and slow waves may be nonspecific, there are reports suggesting that spikes with superimposed or after-going gamma oscillations are more specifically associated with epileptogenic cortex (Engel et al. 2003). Plastic changes are known to occur over time in the sensorimotor cortex (Merzenich and Sameshima 1993). A possible explanation of rapid recovery after limited perirolandic resections may be the presence of a number of functional cortical units within or adjacent to the primary somatosensory cortex that subserve overlapping sensory functions, similar to those which have been described in the motor system (Duffau 2001, Sanes et al. 1995). Such multiple representations of the body in the somatosensory cortex have been found in primates (Tanji and Wise 1981, Wise and Tanji 1981). Indeed, there is a higher propensity of long term potentiation, commonly believed to underlie plasticity and learning (Bliss and Collingridge 1993), after experimental stroke in the sensorimotor cortex of rats (Hagemann et al. 1998), possibly facilitating recruitment of perilesional parallel networks. In adult humans, reorganization of sensory function induced by peripheral or central injury possibly occurs by unmasking nearby latent eloquent sites (Duffau et al. 2000). For example, plasticity in the somatosensory hand area has been demonstrated by magnetoencephalography after hand surgery (Mogilner et al. 1993), and electrocortical stimulation demonstrated functional reorganization of sensory function in cortical areas within and around gliomas (Duffau et al. 2002). Such sites may mediate either the same or different sensory qualities, but with significant overlap to account for recovery of function (Gregorie and Goldring 1984). This may explain why craniotomies with limited resections that spare such representations may yield a favorable outcome (Cohen-Gadol et al. 2003), whereas more extensive resections may be associated with permanent deficits. Thus, chances of remarkable recovery may be enhanced if the excision is guided by the patient’s signs or symptoms in the central sulcus.

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