ERVE damage can lead to a sustained condition referred to as CRPS Type II, formerly referred to as causalgia.31,32 This condition may be associated with a variety of stimulus-evoked signs and stimulus-independent symptoms but usually includes sustained burning pain, allodynia, and hyperpathia combined with vasomotor and pseudomotor dysfunction, and later trophic changes.2,4,18,23,30,32 In some cases of Types I (formerly known as reflex sympathetic dystrophy) and II CRPS, pain and sensory abnormalities have a tendency to spread and eventually to involve the upper quadrant or the entire hemibody.27

Current pathophysiological concepts on central disturbances in CRPS concentrate on abnormalities of pain and sensory processing in the spinal cord;2,4,12 however, more centrally located structures are likely to play an important part in the pathophysiology of CRPS. The hemisensory deficit observed in patients with CRPS may indicate functional disturbances of noxious-event processing in the thalamus.27

Motor cortex stimulation was first proposed for the treatment of central pain and shown to be effective in relieving trigeminal neuropathic pain.19,33,34 Although MCS has been reported to be an effective treatment for peripheral neuropathic pain (for example, brachial plexus avulsion and postherpetic pain), its effectiveness in the treatment of peripheral neuropathic pain caused by CRPS Type II has not been previously reported.6,21,22,28,29

Case Report

History and Examination. This 43-year-old man presented with a severe spontaneous burning pain in his left hand and forearm and allodynia over the left arm and left hemibody. Three years prior to admission, he had experienced a crush injury to his second interdigital space and index finger, and had undergone amputation at the proximal interphalangeal joint of the index finger. After the amputation, a severe burning pain developed in the stump of his ring finger. It was believed, based on diagnostic impression, that this rep-
resisted a digital nerve neuroma. An excision of the neuroma was performed 14 days after the amputation; however, after temporary relief, the burning pain recurred and then spread to the palm and the dorsum of the hand. This pain proved to be intractable and was not relieved by medical treatment or a local anesthetic block. Surgery to excise a neuroma was performed three times on the palm and dorsum of the left hand. The burning sensation, however, was aggravated after a temporary period of relief and was so intolerable that amputation was performed at the level of the second metacarpophalangeal joint. Subsequently, the burning pain extended over the left forearm. Mechanical and coldlike allodynia developed 1 year after the injury and later extended from the forearm to the upper arm and then to his left hemibody including the face, trunk, and lower extremities. Although the patient had undergone repeated neuroma excisions, and there was marked hypesthesia and dysesthesia of the left forearm. Hypesthesia was present on the left hemibody including left hemiface and muscle atrophy in the left hand. His left lower extremity also showed minimal weakness. The distribution of his pain and allodynia are presented in Fig. 1.

Surgical Procedure. Anatomical localization of the central sulcus was performed using a three-dimensional image-guided navigation system. After marking the course of the central sulcus over the vertex, a 5-cm-diameter craniotomy was performed after induction of propofol anesthesia with local infiltration of 1% lidocaine. The surgical procedure performed in this patient was similar to those reported previously. The exact somatotopy of the precentral gyrus was determined using bipolar stimulation with an interpolar distance of 10 mm. After verifying muscle contraction in the painful areas (arm and hand) with a 6- to 7-mA, 1- to 2-Hz electrode array, a four-plate electrode (each separated by 5 mm, diameter 5 mm, model 3587A; Medtronic Inc., Minneapolis, MN) was placed parallel to the mediolateral orientation of the precentral gyrus. During the 3-day test stimulation period, significant pain relief was observed when the 2.5 V, 50 Hz, 450 μsec (“0” negative, “3” positive) combination was used. During stimulation, a sensation of warm paresthesia was produced in the hand and forearm and spread to the upper arm and the ipsilateral neck, face, trunk, abdomen, and the inguinal area. A pulse generator (model 7425; Medtronic) was implanted into the left subclavicular area after induction of anesthesia in the patient (Fig. 2).

Clinical Assessment of Pain Relief. Visual analog scales graduated from 0 to 100 and the McGill Pain Questionnaire were used to assess pain relief. Because peripheral neuropathic pain manifests itself as either spontaneous pain (stimulus-independent pain) and/or as pain hypersensitivity elicited after a stimulus (stimulus-evoked sign or stimulus-dependent pain), we evaluated these symptoms and signs separately. To determine the effect of MCS on the subtypes of allodynia, we applied manual light pressure to the skin to assess mechanical static allodynia, a von Frei hair for mechanical punctate allodynia, and stroking the skin with gauze for mechanical dynamic allodynia. The acetone test was used to examine the effect of thermal cold hyperalgesia. Sensory and motor signs and symptoms of the painful area were evaluated by conventional neurological examination.

Results of Pain Relief. The level of stimulation was adjusted until satisfactory pain relief was achieved by setting the electrode plate at 50 Hz, 2.5 V, and 450 μsec. The post-stimulation analgesic effect was estimated to last 10 minutes, and therefore stimulation was applied continuously in a cyclic mode of 50 minutes on and 10 minutes off.
Motor cortex stimulation for complex regional pain syndrome

The effects of stimulation were evaluated preoperatively and 12 months postoperatively. The visual analog scale score improved from 95 to 10 with stimulation, and the spontaneous burning pain in the left forearm and hand improved by 95%. Changes in the pain scores according to the stimulation parameters are summarized in Tables 1 and 2, which also show the excellent effect of stimulation on various stimulus-independent symptoms and stimulus-dependent signs. Mechanical punctate allodynia and thermal cold hyperalgesia were improved by MCS, and skin temperature over the left forearm increased from 34.1 to 36.2°C; however, allodynia in the skin area without paresthesia did not show the same level of improvement as that with paresthesia. The patient’s pain changed minimally over 12 months of follow up. The mild weakness of the left lower extremity disappeared and the mobility of the left upper arm increased significantly with MCS-produced improvement of pain and allodynia. Unwanted motor contraction and seizure activity have not been observed. After 12 months of stimulation, the patient’s medications could be decreased to 300 mg gabapentin and 30 mg amitriptyline per day.

Discussion

Complex regional pain syndrome is characterized by the presence of regional pain and sensory changes, usually following a traumatic event. Two types of CRPS can be distinguished. Type I, formerly called reflex sympathetic dystrophy, occurs without a definable nerve lesion. Type II, formerly called causalgia, occurs when a definable nerve lesion is present.3,5

Sensory Disturbances in CRPS

Several authors have suggested that pathophysiological mechanisms in the central nervous system play an important part in producing CRPS.2,12 Current pathophysiological concepts concerning central disturbances in CRPS are based on abnormalities of pain and sensory processing in the spinal cord.2 The hemisensory deficit observed in patients with CRPS might indicate functional disturbances in thalamic noxious-event processing.25 Recently Rommel, et al.,7 investigated the extent and quality of sensory impairment and their relation to pain characteristics and movement disorders in patients suffering from CRPS Type I. According to their study, patients with sensory abnormalities in the upper quadrant of the body or a hemisensory impairment experienced predominantly left-sided CRPS (cortical dominance) and demonstrated a significantly higher frequency of mechanical allodynia/hyperalgesia and movement disorders than patients with sensory impairment restricted to a limb. As a possible explanation of cortical dominance, they pointed out the specialization of the right hemisphere for somatic state monitoring, which includes pain.20 Right hemispherical lesions are associated with a syndrome of “neglect,”7,10 and in left-sided CRPS, functional neglect syndrome with decreased perception of the sensory input from the remaining contralateral hemibody might be the consequence of excessive sensory input from the painful area of the affected limb.10 In addition, hemisensory deficit, as observed in their study, had features similar to the sensory disturbances described with large lesions of the contralateral parietal cortex or with lesions in the ventral posterior nuclear group of the contralateral thalamus.3,5

Possible Role of MCS in CRPS With Hemisensory Deficit

The reason we examined MCS was because of the possibility that central nervous system structures such as the thalamus fulfill an important role in the pathogenesis of CRPS Type II, which shows proximal extension of allodynia and sensory disturbance. If this hypothesis is correct, thalamic stimulation and MCS would theoretically be reasonable treatment options. According to reports, thalamic stimulation relieves deafferentation pain in some cases, but electrode implantation is a complicated procedure and its rate of effectiveness is unsatisfactory.11,16,19 Although several retrospective analyses have shown that spinal cord stimulation controls pain in patients with reflex sympathetic dystrophy,1,14,15,26 the effects of spinal cord stimulation in particular patients with CRPS Type II associated with hemibody allodynia and sensory disturbance have not been reported.

Despite encouraging reports on the efficacy of MCS on various types of neuropathic pain, the exact analgesic mechanism of MCS remains a matter of controversy.4,5,13,21 Authors of several studies involving the use of positron emission tomography scanning have indicated that the thalamus is a key structure in the mediation of functional MCS effects.8,9,24,25 Studies on groups of patients with intractable
painless origins have confirmed the existence of significant CBF increases during MCS in the lateral and medial thalamus, the anterior cingulate–orbitofrontal cortex (BA32), the anterior insula–medial temporal lobe, and the upper brainstem near the periaqueductal gray matter. The thalamic areas showing the most significant CBF changes were the ventrolateral and ventroanterior nuclei, that is, the thalamic regions directly connected with motor and premotor cortices. Conversely, no significant CBF changes appeared in the primary motor or somatic areas during the procedure. These results are taken as evidence that descending axons rather than apical dendrites are primarily activated by MCS, and highlight the thalamus as the key structure in the mediation of functional MCS effects. If MCS exerts its analgesic effect through the modulation of thalamic activities, MCS can be applied in particular cases of CRPS with hemisensory deficit, in which functional alterations in the central processing of noxious events may play an important pathogenic role.

Several Singular Stimulation Responses in CRPS Type II

We noted several distinct stimulation responses in this particular case of CRPS. On stimulation at 450 μsec, 50 Hz, 1.5 V, the patient reported that a sensation of warm paresthesia developed initially in the hand and forearm and on increasing the stimulation voltage from 1.5 to 2.5 V, this sensation spread to the upper arm and ipsilateral neck, face, trunk, abdomen, and the inguinal area. In the area covered by warm paresthesia generated by MCS, the spontaneous burning pain was almost completely alleviated and both the mechanical and cold-like allodynia decreased significantly. This stimulation response, however, did not spread further despite increasing the stimulation intensity beyond 2.5 V. This is an epiphenomenon that we had not expected and it has not been previously reported.

The effect of MCS has been reported to be confined to the motor cortex somatotopy. We placed a four-electrode array over the motor cortex corresponding to the hand and forearm somatotopy, because our main goal was to use MCS to relieve the severe burning pain and allodynia over the hand and forearm, the most painful regions. Considering that the spreading of a stimulation response beyond the area of somatotopy has never been reported in other types of central and peripheral neuropathic pain (for example, posther-

### Table 1

**Results of pain relief according to MCS**

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Degree of Improvement (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>stimulus-independent symptoms</td>
<td></td>
</tr>
<tr>
<td>continuous burning pain</td>
<td>95</td>
</tr>
<tr>
<td>intermittent shooting pain</td>
<td>100</td>
</tr>
<tr>
<td>paresthesia</td>
<td>90</td>
</tr>
<tr>
<td>stimulus-evoked signs</td>
<td></td>
</tr>
<tr>
<td>hyperalgesia</td>
<td>70</td>
</tr>
<tr>
<td>mechanical pinprick</td>
<td>80</td>
</tr>
<tr>
<td>thermal cold</td>
<td>50</td>
</tr>
<tr>
<td>allodynia</td>
<td>80</td>
</tr>
<tr>
<td>mechanical static</td>
<td>80</td>
</tr>
<tr>
<td>mechanical dynamic</td>
<td>90</td>
</tr>
</tbody>
</table>

* Cut-off value for von Frei hair.

### Table 2

**Results of mechanical punctate pain threshold weight by using von Frei hair**

<table>
<thead>
<tr>
<th>Part of Body</th>
<th>Off (N)</th>
<th>On (N)</th>
</tr>
</thead>
<tbody>
<tr>
<td>palm</td>
<td>0.028</td>
<td>15.13*</td>
</tr>
<tr>
<td>back of hand</td>
<td>0.068</td>
<td>15.13</td>
</tr>
<tr>
<td>forearm</td>
<td>0.69</td>
<td>15.13</td>
</tr>
<tr>
<td>trunk</td>
<td>0.69</td>
<td>15.13</td>
</tr>
<tr>
<td>leg</td>
<td>1.02</td>
<td>15.13</td>
</tr>
</tbody>
</table>

* Cut-off value for von Frei hair.

### Conclusions

Motor cortex stimulation proved to be effective in this particular patient with CRPS Type II, in whom hemibody alldynia and sensory disturbances also occurred. Although it is difficult to explain, MCS induced stimulation-evoked sensation in the painful area and this sensation spread beyond the somatotopy of the motor cortex where the electrode was placed. As shown by this case, a central mechanism seems to play an important role in the pathogenesis of hemibody sensory disturbance in CRPS. Considering that these conditions are notoriously treatment resistant, we believe that MCS may be a reasonable option for the management of this kind of peripheral neuropathic pain. The application of MCS for the treatment of pain confined to the involved extremities in CRPS without hemibody sensory involvement should be approached cautiously. Further detailed studies are needed to determine the pathogenesis of CRPS and the analgesic effects of MCS.

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References


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