Improving writer’s cramp dystonia after prolonged muscle stimulation. Report of two cases

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Abstract
Focal primary dystonia has been recognized as a motor disorder; nevertheless, some studies suggest that sensory dysfunction might be involved. We report two patients who improved their writer’s cramp dystonia after deep muscle stimulation using acupuncture needles. We support the hypothesis that deep and prolonged stimulation of muscles related to dystonia can modify the cortical sensory-motor response and improve writer’s cramp.

Keywords: Writer’s cramp, Deep muscle stimulation, New approach.

Resumen
La distonía focal primaria ha sido reconocida como un trastorno motor. Sin embargo, algunos estudios sugieren que una disfunción sensorial puede estar involucrada. Nosotros reportamos dos casos clínicos que corresponden a pacientes que mejoraron su distonía de calambre del escribiente después de la estimulación muscular utilizando agujas de acupuntura. Nosotros apoyamos la hipótesis de que la estimulación profunda y prolongada de los músculos relacionados con la distonía pueden modificar la respuesta sensitivo-motora cortical y mejorar el calambre del escribiente.

Palabras Clave: Calambre del escribiente, Estimulación muscular profunda, Nuevo enfoque.

Introduction
Focal primary dystonia (FPD) has been recognized as a motor disorder, although clinical, neurophysiological and imaging observations suggest that sensory dysfunction might also be involved.1-4

Underlying mechanisms that involve frequent, repetitive and temporarily related stimuli could be a factor triggering focal dystonia. It has been shown that the primary sensory cortex (S1) and secondary parietal cortex record increased activity during dystonic posture.5 Some studies have found that, in patients with focal hand dystonia, there is an abnormal representation in S1 and that this abnormality may be related to the severity of the dystonia. 5 In these patients, there may be a loss of the cortical inhibition, and an increase in neuronal plasticity.6

We report two patients with writer’s cramp dystonia who responded well to prolonged and repeated stimulation of their hand muscles with acupuncture needles.

Patient 1
Sixty-year-old, right-handed male patient, writer by profession, with a right writer’s cramp dystonia that started four years earlier. One year before he had been administered a treatment with botulinum toxin, which improved the dystonia for six months.

We performed a muscle puncture in his right hand using acupuncture needles in the abductor pollicis brevis and first interosseus dorsalis muscles for 30 minutes.

After this procedure, the patient showed significant improvement in his writing for more than 72 hours. The procedure was repeated twice a week for a month and then once a week. The patient has shown a significant improvement until one year after of the first puncture.

Patient 2
Seventy-two-year-old, right-handed male patient, lawyer, with a writer’s cramp dystonia that started five years earlier. He had not been able to write for 30 years. He had not been able to write for a year before treatment (Figure 1a). We performed a muscle puncture in his right hand using acupuncture needles in the abductor pollicis brevis and first interosseus dorsalis muscles for 30 minutes. Immediately after the first muscle puncture the patient was able to write again with mild difficulty (Figure 1b). The procedure was repeated twice a week for a month and then once a month for six months.

The patient has been able to return to his previous work as a writer. The patient has continued working until one year after of the first stimulation with acupuncture needles (Figure 1c). The patient did not take any medication for dystonia during treatment.

Discussion
Our results show that writer’s cramp dystonia may improve with deep repeated muscle stimulation in the abductor pollicis brevis and first interosseus dorsalis, with the consequent mitigation of its severity and persistence over time.7 Despite the evident motor manifestation of idiopathic focal hand dystonia, it has been recognized that the sensory system performs an important role in this condition.1,2,3,4,8 There is much evidence substantiating sensory dysfunction in patients with dystonia: sensory symptoms may precede the appearance of dystonia; certain sensory tricks used by patients may help to relieve dystonic postures and sensory training such as the one used for Braille reading5 and peripheral blockage may relieve the dystonic posture of the hand.9,10 Sensory dysfunction may contribute to a loss of sensory-motor integration and abnormality of the motor output in focal dystonia.1,5,11

The sensory system is a major drive for the motor system and the basal ganglia perform an important role in the central processing of the somato-sensory drive.4,5,11. The temporal discrimination, a function of the basal ganglia, and the spatial discrimination, a function of S1, are impaired in dystonia.

The sensory dysfunction, however, if not the primary event in dystonia, may certainly contribute to impaired sensorimotor integration and abnormal motor output.5
In patients with focal hand dystonia, various abnormalities have been shown at different levels of sensory processing. In experiments with animals, sensory studies with highly repetitive stimuli induce an abnormal process of sensory information and a remodeling of the neuronal plasticity of the primary somato-sensory cortex. Imaging studies of patients with writer’s cramp indicate an increased output from the basal ganglia to the thalamus and the cortical areas, with an upregulation of the pre-motor and motor cortex with co-contraction and dystonic posture. Studies with fMRI in patients with focal hand dystonia have shown an abnormality in the cortical representation of individual digital in the primary somato-sensory cortex. In these patients, the cortical representation of the fingers of the hand is compressed and disorderly. Increase and overlay of the receptive fields have been found, as well as an occasional inversion of the digital representation and a loss of the cortical inhibitory function, probably with fluctuation in the activity of inter-neurons in S1 and M1. The hypersensitivity and expansion of the cortical sensory-motor representation corroborates the hypothesis that dystonia is a sensory disorder produced by an environmental experience. Neurophysiological research has revealed cortical abnormalities in patients with focal dystonia that reflect an underlying genetic component.

Our results suggest that prolonged and repeated stimulation of certain muscles of the hand related to writer’s cramp produce a favorable therapeutic response that persists over time, this contributes to demonstrating the sensory participation of dystonia but also shows that intense peripheral sensory input could achieve modification of the cortical sensory-motor response.

We do not know about the neurophysiological mechanisms whereby intense and prolonged stimulation of the muscle is capable of producing a therapeutic response that persists over time. It is probably due to a modification related to neuronal plasticity of the cortical inter-neurons. Our findings open up new avenues of physiopathological and therapeutic research.

**Bibliografía**