Clinical Neurophysiology 118 (2007) 2215–2226

www.elsevier.com/locate/clinph

Abnormal spinal interactions from hand afferents to forearm muscles in writer's cramp

G. Lourenço [a,b], J.-P. Bleton [a,b,c], C. Iglesias [a,b], M. Vidailhet [d,e,f], V. Marchand-Pauvert [a,b,*]

[a] *INSERM, U731, Paris F-75013, France*
[b] *Université Pierre et Marie Curie-Paris 6, UMRS-731, Paris F-75005, France*
[c] *Hôpital Sainte Anne, Neurologie, Paris F-75014, France*
[d] *INSERM, U679, Paris F-75013, France*
[e] *Université Pierre et Marie Curie-Paris 6, UMRS-679, Paris F-75012, France*
[f] *AP-HP, Hôpital Saint Antoine, Neurologie, Paris F-75012, France*

Accepted 5 July 2007

Abstract

Objective: Spinal reflexes from hand to wrist muscles were investigated in writer's cramp.
Methods: Stimulus-triggered rectified EMG averages after ulnar nerve and cutaneous stimulation, in wrist flexors and extensors during tonic contraction, were compared in 18 controls and 19 patients.
Results: On the patient dystonic side, ulnar-induced EMG suppression was decreased in wrist extensors, and facilitation in wrist flexors modified dependent on the dystonic wrist posture during writing. No change was found on the patient non-dystonic side. Cutaneous stimulation increased wrist flexor EMG on both sides of the patients with normal wrist posture during writing, but had no effect in controls and patients with abnormal wrist posture.
Conclusions: Comparison between cutaneous and mixed nerve stimuli suggests that spindle afferents from intrinsic hand muscles may mediate patients' ulnar-induced EMG modulations. Abnormal proprioceptive control was only observed on dystonic side, while bilateral unusual cutaneous control was found in patients. Changes in spinal transmission were partly related to the dystonic wrist posture, suggesting that systems involved in sensory processing can be differentially altered in writer's cramp.
Significance: Changes in spinal transmission, probably related to peripheral and/or cortical inputs, might either take part in primary or adaptive mechanisms underlying writer's cramp.

Keywords: Dystonia; Human; Motor control; Spinal reflex; Sensory inputs; Propriospinal

1. Introduction

Dystonia is defined by abnormal movements and postures, interfering with motor performance. Writer's cramp is a focal, task-specific dystonia, manifested only during handwriting tasks. Both genetic (Defazio et al., 2003a,b) and environmental factors (Byl et al., 1996) may be involved in the pathophysiology of dystonia, but understanding the neurophysiological mechanisms underlying writer's cramp is made difficult given the heterogeneity of results from brain imaging and electrophysiological studies. Loss of central inhibition and abnormal sensory functions are nevertheless well characterised in writer's cramp, at both cortical and sub-cortical levels (see Hallett, 2006). However, it is difficult to determine if those changes are endophenotypic markers of focal hand dystonia and/or compensatory changes. It is usually suggested that primary

* Corresponding author. Address: U731 INSERM/UPMC – Physiologie et Physiopathologie de la Motricité chez l'Homme, S[ce] MPR, Hôpital Pitié-Salpêtrière, 47 bd de l'Hôpital, 75651 Paris cedex 13, France. Tel.: +33 1 42 16 11 20; fax: +33 1 42 16 11 02.
E-mail address: veronique.marchand@chups.jussieu.fr (V. Marchand-Pauvert).

1388-2457/$32.00
doi:10.1016/j.clinph.2007.07.009

mechanisms should affect both dystonic and non-dystonic sides, and compensatory effects would only be manifested on the dystonic side.

At the spinal level, the short-latency response to muscle stretch is unmodified in writer's cramp (Rothwell et al., 1983; Tatton et al., 1984; Matthews, 1991). However, spinal inhibition between wrist flexors and extensors is decreased although non-consistent results have been observed for reciprocal group Ia inhibition (no change, Nakashima et al., 1989; decrease on the dystonic side, Panizza et al., 1989; decrease on both sides, Chen et al., 1995). On the other hand, if presynaptic inhibition of group Ia terminals is decreased in all studies, these changes are not specific to dystonia (Nakashima et al., 1989). Finally, median- and radial-induced modulations of EMG activity in wrist flexors and extensors have been found reduced in these patients with respect to controls (Valls-Sole and Hallett, 1995).

All these results concerned either homonymous responses (i.e. muscle response to stimulation of its own afferents) or response to stimulation of the nerve supplying antagonistic muscles acting at the same joint. Heteronymous spinal reflexes evoked by stimulating hand muscles afferents onto various forearm muscles have been recently revealed in healthy human subjects while investigating the effects of ulnar nerve stimulation at wrist level on (i) single motor unit discharge, (ii) changes in monosynaptic reflex amplitude, and (iii) post-stimulus-triggered rectified EMG averages (Lourenço et al., 2006, 2007). Changes reported in wrist flexor motoneurones included early non-monosynaptic group I excitation mediated through excitatory propriospinal-like interneurones (Lourenço et al., 2006; Iglesias et al., 2007; see Pierrot-Deseilligny and Burke, 2005), and late excitation transmitted within its first 4.5 ms through group II spinal pathway (middle phase; Lourenço et al., 2006). Changes in wrist extensor motoneurones were a massive inhibition, possibly mediated via inhibitory propriospinal-like interneurones (Lourenço et al., 2007).

Spinal reflexes from hand muscles afferents to motoneurones supplying wrist muscles are supposed to stabilise normal wrist posture during hand movements (Lourenço et al., 2007). Given the possible abnormal motor coordination between hand and wrist muscles, and the resulting dystonic hand postures in focal hand dystonia, we investigated whether transmission in these pathways could be modified in patients with writer's cramp. Since abnormal cutaneous control has been reported in writer's cramp at the cortical level (Meunier et al., 2001), we also investigated the effects of purely cutaneous stimuli, which are only weak and rarely significant in healthy subjects (Lourenço et al., 2006, 2007).

2. Methods

The experiments were performed on 18 control subjects (23–71 yr; mean 39.9 ± 3.4; 10 females), and 19 patients with writer's cramp (19–67 yr; mean age 46.4 ± 3.6; 9 females); all wrote with the right hand. Subjects' consent was obtained according to the Declaration of Helsinki, and the Ethical Committee of the Pitié-Salpêtrière Hospital approved the experimental procedures.

2.1. Patient description

The patients suffered from unilateral simple cramp only triggered during handwriting. None of them had other focal dystonia affecting other parts of the body, or had started writing with the left hand. The age of cramp onset was 38.7 ± 3.6 yr, and the duration of symptoms, 8.3 ± 2.2 yr. The sub-score for handwriting on the Dystonia Disability Scale was 1.6 ± 0.2 (range 1–4; 0 for normal writing, 1 for slight difficulties, 2 for almost illegible, 3 for illegible, and 4 for unable to hold the pen; see Burke et al., 1985). Tests were done before commencing rehabilitation, and no patient had been taking medication or had received botulinum toxin injection. Despite the difficulty of analysing dystonic movements during handwriting, 3 subgroups of patients could be distinguished: those without dystonic wrist postures, those with predominant dystonic wrist posture of inflexion, and those with predominant dystonic wrist posture of extension during handwriting. The 3 groups were established during the clinical evaluation, before protocol inclusion and data analysis.

2.2. Recordings

Subjects were seated with the forearm in the prone position. The right side was investigated in all 37 subjects, and the left side was explored in 18 of them (9 controls and 9 patients). The electromyogram (EMG) was recorded by surface electrodes (two 0.5-cm^2 silver plates; 1.5 cm apart) secured to the skin over Extensor Carpi Radialis (ECR), and Flexor Carpi Radialis (FCR). The level of rectified and averaged EMG produced during the maximal voluntary tonic contraction (MVC), sustained during 1 min, was first estimated in each subject. As already reported by Valls-Sole and Hallett (1995), there was no difference between controls and patients (respectively, 105.8 ± 15.2 vs. 96.5 ± 16.7 μV in FCR, $P = 0.68$; 77.1 ± 15.1 vs. 94.9 ± 16.4 μV in ECR, $P = 0.43$; unpaired Student's t-test). During recordings, subjects were asked to perform steady tonic contractions of 20% of the maximal EMG activity (recorded during MVC) with the help of visual feedback (rectified and averaged EMG was displayed on an oscilloscope). Patients were able to maintain this level of 20% without exhibiting dystonic movement or abnormal co-contraction of wrist flexors and extensors during the tests. EMG was filtered (0.1–1 kHz), amplified (×10,000), rectified, and digitally recorded using a 2-kHz sampling rate.

2.3. Conditioning stimuli

Conditioning consisted of percutaneous electrical stimuli (1-ms rectangular pulse; two 0.5-cm^2 silver plates;