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Improved identification of dystonic cervical muscles via abnormal muscle activity during isometric contractions

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ABSTRACT

Background: The preferred treatment for cervical dystonia (CD) is injection of botulinum toxin in the dystonic muscles. Unfortunately, in the absence of reliable diagnostic methods it can be difficult to discriminate dystonic muscles from healthy muscles acting in compensation. We investigated if dystonic muscle activation patterns could be identified in cervical dystonia patients during a harmonized isometric contraction task. Furthermore, we investigated whether dystonia worsens at higher levels of voluntary contraction, which might further improve the identification of dystonic muscle activity.

Methods: An isometric device was used to investigate muscle activation during voluntary contraction tasks in 10 controls and 10 CD patients. Surface electromyography (EMG) of the sternocleidomastoidus, splenius capitis, and semispinalis capitis muscles was evaluated during a rest task and when performing submaximal (20%) and maximal voluntary contractions for eight head transversal force directions and for head twist. Two measures were developed to identify dystonic activation: 1) Muscle activity in the contraction direction in which the contribution of the muscle was lowest (Minimum EMG), and 2) the average muscle activity over all contraction directions (Total Mean EMG).

Results: Patients showed increased dystonic activity in the rest task and during submaximal contractions relative to controls, but not during maximal contractions. Increases in Minimum EMG indicated an inability of patients to deactivate dystonic muscles counteracting the task. Increases in Total Mean EMG indicated dystonic activity in all task directions. During maximal contractions these effects were absent in dystonic muscles. Dystonia is therefore found not to worsen at higher levels of isometric voluntary contraction. The activity of dystonic muscles modulated with different loading directions similar to controls. Using Minimum EMG 54% of the muscles clinically diagnosed as dystonic and 91% of non-dystonic muscles were predicted correctly.

Conclusions: Dystonic muscle activity was found in cervical dystonia patients during submaximal contractions in all task directions using a harmonized isometric task, but no differences were found during maximal contractions. With some adaptation this method may prove useful to identify dystonic muscles.

1. Introduction

Cervical dystonia (CD) is a movement disorder of the neck characterized by involuntary activity of neck muscles leading to debilitating abnormal postures and twisting movements and pain [4,5]. Conventional methods to identify dystonic muscles for treatment with botulinum toxin (BoNT) involve palpation and analysis of head posture [11,12]. However, in many patients it remains difficult to discriminate dystonic muscles from healthy muscles acting in compensation [2,25]. Consequently, inadequate muscle selection might be an important cause of therapy failure [7,24]. A number of studies [11,27,17] propose electromyographically-guided BoNT injections as a systematic approach in the identification of dystonic muscles, where levels of muscle activity are evaluated when patients are

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asked not to resist their dystonic posture. However, even then it remains unclear whether increases found in EMG are due to dystonic activity, or due to compensatory muscle activation. EMG studies for dystonic muscle identification are generally performed when patients are asked to minimize volitional muscle contractions and allow the dystonic posture to go freely [11,27]. On the other hand, different studies found dystonic muscle activity to worsen when patients performed voluntary actions [1,15,20] suggesting that dystonic muscle activity may best be identified when the muscle contracts. For this study, an isometric device was developed that enabled us to evaluate neck muscle activity during isometric voluntary contractions in different directions. This method was used to investigate muscle activation during standardized isometric tasks in controls and CD patients. Because subjects were comfortably fixed with their head in the isometric device, the need for muscles to compensate for dystonic movements was minimized. The goal was to identify dystonic muscle activation in different task directions and at varying levels of voluntary muscle contraction. We hypothesized that during these isometric experiments compensatory activity was minimized in healthy muscles and increased EMG activity (i.e., dystonic) could more reliably be identified in patients. Within patients, we investigated the correlation between muscles showing dystonic activation patterns and the clinically dystonic muscles. Furthermore, we hypothesized that at higher levels of voluntary contraction dystonic activity would worsen, which would further improve the identification of aberrant muscle activity.

2. Subjects and methods

2.1. Ethical standards

The experimental protocol was in accordance with the Declaration of Helsinki and was approved by the ethics committee at the Delft University of Technology and the medical ethics committee at the Amsterdam Medical Centre. All subjects gave written informed consent.

2.2. Subjects

The experiments were performed with ten CD patients (five males, age 56 ± 11 years) and ten age matched controls (four males, age 55 ± 14 years). The TSUI [26] and TWSTR (Toronto Western Spasmodic Torticollis Scale) [6] scales were used to quantify the severity of the disorder. All patients were treated with botulinum toxin (BoNT) with the last injection provided at least three-months prior to the experiment. For the purposes of this study, muscles that were injected with BoNT during clinical treatment shortly after this experiment were considered to be dystonic. All other patient muscles were classified as unaffected patient muscles. A comparison was subsequently performed between the dystonic, unaffected patient, and healthy control muscles. Dystonic muscles varied per patient and are listed in Table 1. The authors are aware that muscles may have been wrongly categorized as dystonic and address this issue in the discussion.

2.3. Protocol

2.3.1. Apparatus and data collection

Subjects were seated and fixed in an isometric device with a tightly fitted cushioned helmet. They performed neck muscle contraction tasks to generate horizontal (transversal) forces and twist (spinal axial rotation) moments that were measured by an overhead six axis load cell (MC3-6-500, AMTI Inc., Watertown, USA). Force and moment signals were sampled at 2000 Hz. Surface EMG was recorded bilaterally with paired unipolar electrodes (TMS International BV, Oldenzaal, The Netherlands) placed over sternocleidomastoid (SCM), splenius capitis (SPL), and semispinalis capitis (SS) muscles at a sample rate of 2000 Hz. Subjects received visual feedback of the applied transversal force and twist moment to ensure effective task performance. Visual feedback was presented through a custom made interface using Matlab (Matlab R2011b, Mathworks Inc., Natick, MA, USA) (Fig. 1).

In some patients tremulous head movement was observed. The level of dystonic tremor [9] present in the subjects was therefore evaluated in a separate secondary experiment. Subjects were fixed in a chair with the head free to move, and three-dimensional motion of six markers on the head was recorded at 200 Hz using an Oqus 6-camera motion capture system (Qualisys AB, Gothenburg, Sweden), following similar methods described in a previous study [18].

2.3.2. Task instruction

The primary experiment consisted of three isometric tasks, each with a different level of muscle contraction in order to assess the effect of muscle activation levels on dystonic muscle behavior. In the first task, subjects performed maximum voluntary muscle contractions (MVC) using neck muscles to generate head forces in eight transversal directions at 45° intervals, and maximum head moment in left and right axial rotation. In the second task, subjects performed submaximal voluntary contractions (20% MVC) in the same directions as the MVC trials. MVC trials were repeated three times lasting 5 s each and 20% MVC trials were repeated two times lasting 10 s each. Subjects were

Table 1
Patient characteristics. The TSUI and TWSTR scales indicate the severity of the disorder. The TSUI score observes the amplitude and duration of sustained movements and tremors with a maximum score of 25. The TWSTR scale adds the subject's severity of Torticollis (0–35), disability (0–30), and pain (0–20) and has a maximum score of 85. High values always indicate an increased severity. The dystonic sternocleidomastoid (SCM), splenius capitis (SPL), and semispinalis capitis (SS) are given per patient based on treatment. Tremor has been identified in patients 2, 7, and 8 during this experiment.

#	Gender	Age	Information on disorder						Muscles treated with BoNT					
			TSUI	TWSTR	Duration (years)	Rot L[b]	Rot R	Tremor or jerk[c]	SCML (6)[a]	SPLL (4)	SSL (3)	SCMR (4)	SPLR (7)	SSR (4)
1	M	38	7	16	4.5		2		1				1	
2	F	63	11	30.1	4		2	1	1	1			1	
3	F	56	9	41.75	4		3		1				1	
4	M	46	8	23	11		1		1			1		
5	F	74	12	24	25	3				1	1	1		1
6	M	45	16	22	4		3		1		1		1	
7	F	63	17	31	6	2		1		1		1		1
8	F	60	15	24	10	2		1		1	1		1	1
9	M	56	7	17	13		1		1				1	
10	M	61	13	29.75	2	3				[d]		1	1	1

BoNT. botulinum toxin; TWSTR. Toronto Western Spasmodic Torticollis Rating Scale; Rot L. rotation left; Rot R. rotation right; SCML. left sternocleidomastoidus; SPLL. left splenius capitis; SSL. left semispinalis capitis; SCMR. right sternocleidomastoidus; SPLR. right splenius capitis; SSR. right semispinalis capitis.

[a] The numbers of dystonic muscles are given in brackets.

[b] Rotation. (−) No deviation. (1) <15°. (2) 15°–30°. (3) >30°.

[c] (−) No tremor. (1) tremor or jerk observed during experiment.

[d] Removed as electrode had come loose during testing.