Visual Feedback Reduces Co-contraction in Children With Dystonia

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Visual Feedback Reduces Co-contraction in Children With Dystonia

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Abstract

Inappropriate muscle activation and co-contraction are important features in childhood dystonia, and clinical interventions are often targeted to reduce the excess muscle activation. Previous research has shown that visual biofeedback of muscle activity can help people to reduce excess muscle activation in a variety of motor disorders. To investigate the effectiveness of similar techniques for dystonia, we had participants perform a tracking task with and without visual feedback of co-contraction. Children with dystonia had greater levels of co-contraction than children without dystonia. Most importantly, individuals were able to reduce their co-contraction significantly when visual biofeedback was provided. These results indicate that children with dystonia are able to control co-contraction, at least to a certain extent, provided attention can be directed to the excess muscle activation. These results also suggest that methods of biofeedback focusing on inappropriate muscle activations might provide a clinical benefit for treatment of children with dystonia.

Keywords

dystonia, electromyography feedback, co-contraction, biofeedback

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Childhood dystonia is defined as "a movement disorder in which involuntary sustained or intermittent muscle contractions cause twisting and repetitive movements, abnormal postures, or both."[1] Dystonia is one of the impairments noted in children with cerebral palsy, brain injury, and metabolic disorders, but it can also have a genetic cause or present as an idiopathic condition. Current interventions for dystonia include physical and occupational therapies; pharmacological approaches; and deep brain stimulation for the most involved cases. Most treatment options, however, address only the symptoms, and they are often not successful in controlling dystonia.[2] As a result, there is a need to investigate alternate options for control of dystonia in children.

One therapeutic approach that has shown promise in several motor disorders is biofeedback. In general terms, biofeedback is the use of instrumentation to make individuals aware of a physiological process of which they usually have very little knowledge.[3] In disorders of movement, this usually takes the form of providing individuals with visual or auditory feedback of muscle activity. By bringing an individual's attention to the covert physiological activity, the individual can learn to develop conscious awareness, and eventually control, over that activity.[4] This approach has been shown to help individuals with a variety of neuromuscular disorders to increase or decrease levels of muscle activity.[3]

Although biofeedback of muscle activity has not been investigated specifically for the treatment of childhood dystonia, studies have shown that it can have positive effects in related disorders. For example, earlier studies have shown positive effects in reducing muscle activation in individuals with athetoid cerebral palsy.[5-7] Researchers have also used biofeedback to help individuals decrease muscle tone in adult focal hand dystonia,[8] spasmodic torticollis,[4] and generalized dystonia.[9] Several studies have shown biofeedback to be helpful for controlling spasticity of the lower limbs in children with cerebral palsy.[10,11] Biofeedback has also been shown to help children with and without traumatic brain injury to reduce muscle overflow between limbs.[12,13]

It is currently not known whether biofeedback approaches can help children with dystonia to control their dystonic symptoms. To help answer this question, the present study focuses

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Table 1. Characteristics of Participants With Dystonia

Participant	Age, y	Gender	Diagnosis	BAD Scale score for the tested arm
D1	8	Male	Striatal necrosis from hemolytic-uremic syndrome	3
D2	9	Female	Attention deficit disorder and torticollis, right writer's cramp	1
D3	10	Female	Right hemiplegic cerebral palsy	3
D4	11	Female	Spastic diplegic cerebral palsy, premature birth	2
D5	11	Female	Right suprasellar dermoid tumor resection	3
D6	11	Male	Left basal ganglia cavernous malformation resection	3
D7	13	Male	Triplegic cerebral palsy	0
D8	13	Female	Dyskinetic cerebral palsy	3
D9	14	Female	Non-*DYT1* primary generalized dystonia	1
D10	14	Female	Diplegic cerebral palsy	2
D11	16	Male	Tetraplegic cerebral palsy	3
D12	16	Male	Left hemiplegic cerebral palsy	4
D13	17	Male	Right hemiplegic cerebral palsy	3
D14	18	Female	Dyskinetic cerebral palsy	4

Note: BAD Scale, Barry-Albright Dystonia Scale.

on co-contraction, the simultaneous contraction of agonist and antagonist muscles. Co-contraction is traditionally considered to be one of the physiological hallmarks of dystonia,[1,14,15] potentially underlying several observed abnormalities of passive and active movement. At the same time, more recent results have suggested that co-contraction cannot always be an obligate feature of dystonia in children.[16]

To determine whether children with dystonia could control their co-contraction with the help of feedback, we had participants perform an isometric task using electromyography signals from their biceps and triceps muscles. The task required alternate contraction of biceps and triceps in a moderated way to track a target on a computer monitor, and we measured the amount of co-contraction between those muscles during the task. To determine the effect of feedback, we provided participants with an indicator of co-contraction during 1 block of trials, and we measured whether this resulted in reduced co-contraction. We also measured tracking error, to ensure that participants did not alter their co-contraction at the expense of tracking performance. The results provide evidence that children with dystonia are able to control co-contraction if they are provided with visual feedback of muscle activity.

Methods

Participants

Participants consisted of a dystonic group containing 14 children with dystonia (8 girls; ages 8–18 years, mean 12.9 years, standard deviation 3.1 years), and a control group containing 37 neurologically healthy children without dystonia (14 girls; ages 6–17, mean 10.8 years, standard deviation 3.2 years). Individuals in the dystonic group were recruited from the Stanford Medical Center and diagnosed by a pediatric neurologist (T.D.S. or Allison Przekop), based on history and clinical examination, according to standard definitions.[1] All participants with dystonia were rated with the Barry-Albright Dystonia Scale.[17] Table 1 provides individual information on all participants in the dystonic group.

Stanford University's Institutional Review Board approved the study protocol, and the study was registered with clinicaltrials.gov (NCT00285870). All participants or their parents gave informed written consent for participation and authorization for use of protected health information.

Apparatus

Participants sat in an adjustable chair (System 2 chair, Biodex Medical Systems, USA) with straps holding their trunk steady. For each participant, we chose a single arm for testing. For the dystonic group, we tested the arm that exhibited the greatest amount of dystonia, and for the control group, we tested the dominant arm. We immobilized the tested arm in a rigid device that held the shoulder abducted 90 degrees and the elbow flexed 90 degrees, with the forearm oriented vertically. Previous publications from our laboratory provide a photograph of the apparatus.[18,19]

Surface electromyography electrodes (DE-2.3, Delsys Inc., USA) with a band-pass filter of 20 to 450 Hz and an amplification of 1000 times were placed over the bellies of the biceps and triceps muscles. The electromyography signals were sampled at 1 kHz using an analog to digital interface (Power 1401, CED Technologies Inc., UK) in connection with custom data acquisition software.

We obtained isometric muscle activation signals for the experimental task by filtering the electromyography signal from each electrode through 3 steps. Each signal was processed with a high-pass Butterworth filter (4th-order, 1 Hz cutoff), then a Bayesian filter, and finally a low-pass Butterworth filter (2nd-order, 5 Hz cutoff). The Bayesian filter produces a smooth output that estimates the driving force underlying the electromyography signal, while also allowing fast low-latency changes in the filtered signal.[20]

Before the start of the experiment, we measured the isometric maximal voluntary contraction for the biceps and triceps muscles. The electromyogram from each electrode was displayed as visual feedback for the participant. The participant performed 3 attempts of 10 seconds of maximum contraction for each muscle. Maximum voluntary contraction was quantified by the data acquisition software as the maximum mean electromyogram measured over a 200-millisecond period during activation.

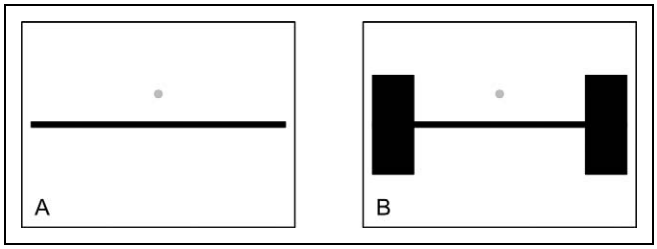

Figure 1. Tracking task used in this experiment. The circle acted as a target, moving vertically with a randomly distorted sinusoidal motion. Participants tracked the target with the horizontal bar (cursor), activating their biceps to move the cursor upward and their triceps to move it downward. (A) Display for Blocks 1 and 3 contained only the target and cursor. (B) Display for Block 2 also included enlargements on each side of the cursor, indicating the amount of co-contraction between the biceps and triceps muscles.

Procedure

Each participant attended a single experimental session of approximately 1-hour duration. The session started with seating at the apparatus, placement of the electromyography electrodes, and measurement of maximum voluntary contraction values. Following this, the participants completed a series of trials during which they tracked a target on a computer screen by activating their biceps and triceps muscles to move a cursor.

During each trial, participants watched a computer monitor displaying a target and cursor, as shown in Figure 1. The target moved vertically with a randomly distorted sinusoidal motion. Target motion was centered on the vertical midline of the screen. Participants tracked the target with the cursor by activating their biceps and triceps muscles. Biceps activity moved the cursor upward, and triceps activity moved the cursor downward. Activation of either the biceps or triceps muscle at 20% of maximum voluntary contraction resulted in the cursor moving to the top or bottom of the screen, respectively. Activation of both muscles resulted in the summation of their respective actions, so that when both muscles were at rest or equally active, the cursor remained at the middle of the screen.

In some trials, participants were also provided with a visual display of co-contraction between their biceps and triceps muscles. Co-contraction was defined as the lesser of the normalized biceps and triceps activity, whereas normalized activation is defined as the ratio of the activation of each muscle to its maximum voluntary contraction (see Malfait and Sanger[16] for a discussion of co-contraction definitions). Co-contraction was displayed as a pair of dumbbell-shaped enlargements, 1 on each side of the cursor, as shown in Figure 1B. The height of the enlargements was linearly related to the amount of co-contraction. Therefore, movement of the cursor without increasing the height of the enlargements could only be achieved by activating 1 muscle while maintaining the antagonist completely relaxed.

Before the start of testing, participants were allowed to practice moving the cursor on the screen for 1 minute. During this time, the experimenter monitored their performance and made adjustments or suggestions to ensure that each participant was able to control the cursor adequately using their arm muscles.

Following the practice trial, each participant performed 12 test trials, arranged in 3 blocks of 4 trials. All trials had a duration of 60 seconds. Participants were given a 30-second rest between trials, and a 60-second rest between blocks. Participants were also able to indicate if they were tired and desired a longer rest.

During the first and third blocks of trials, the monitor showed only the target and cursor, with no indication of co-contraction, as shown in Figure 1A. Participants were instructed to track the target while keeping their arm as relaxed and loose as possible. During the second block, the monitor showed the target, cursor, and the co-contraction indicators, as shown in Figure 1B. Participants were instructed to track the target as the first priority, while also trying to keep the co-contraction indicators as small as possible. They were instructed that the co-contraction enlargements indicated that both of their muscles were active at the same time, and they could reduce the size of the enlargements by relaxing their arm.

Analysis

Co-contraction was defined as the lesser of the normalized biceps and triceps electromyograms. To determine whether participants reduced their co-contraction by reducing movement altogether or by neglecting their tracking performance, we also investigated changes in tracking error. We defined tracking error as the absolute difference between the target position and the cursor position, expressed in units of normalized electromyogram.

To avoid any learning effects associated with starting new blocks or trials, we analyzed only the last 30 seconds of trials 2 through 4 in each block. For each trial analyzed, we used the mean of all samples in the last 30 seconds to obtain single measures of co-contraction and tracking error for that trial. We used a mixed-effects model to test each dependent variable for fixed effects of group, block, and trial, while considering participant as a random factor. Analysis was performed using the lme function from the nlme package[21] of the R statistical computing environment.[22] To correct the skew and heterogeneity of variance seen in the data, we logarithmically transformed the co-contraction and tracking error values before performing the tests. When evaluating each model, we used a criterion for significance of $P < .05$, and we removed nonsignificant components from the model using backward elimination.

Results

Figure 2 shows the mean co-contraction across all conditions for each group. Testing for the effects of group, block, and trial showed no significant 2- or 3-way interactions among the factors. Additionally, trial was not a significant factor, suggesting that co-contraction did not change significantly between trials within each block. Group was a significant factor ($F(1,49) = 6.31$; $P = .015$), indicating that the dystonic group had significantly more co-contraction than the control group. Block was also a significant factor ($F(2,404) = 17.9$; $P < .001$), and tests indicated that blocks 2 and 3 both had lower co-contraction than block 1 ($t(404) < 5.13$; $P < .001$).

Figure 3 shows the mean tracking error across all conditions for each group. Testing for the effects of group, block, and trial showed no significant 2- or 3-way interactions among the factors. Trial was also not a significant factor, indicating that tracking error did not significantly change between trials within each block. Group was a significant factor ($F(1,49) = 11.6$; $P = .001$), indicating that the dystonic group had significantly more error than the control group. Block was also a significant factor ($F(2,404) = 6.45$; $P = .002$). In this case, tracking error was not significantly different in block 2 than in block 1 ($t(404)$

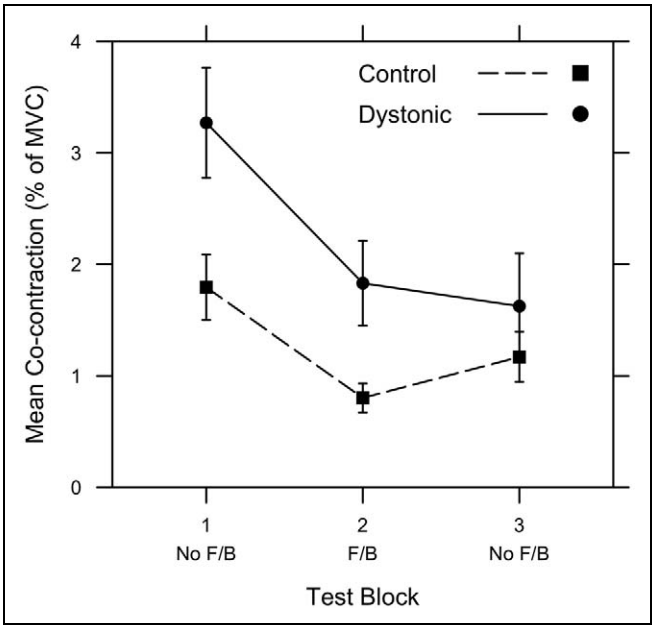

Figure 2. Mean co-contraction for each group and block. Error bars indicate standard errors. MVC, maximum voluntary contraction; F/B, feedback.

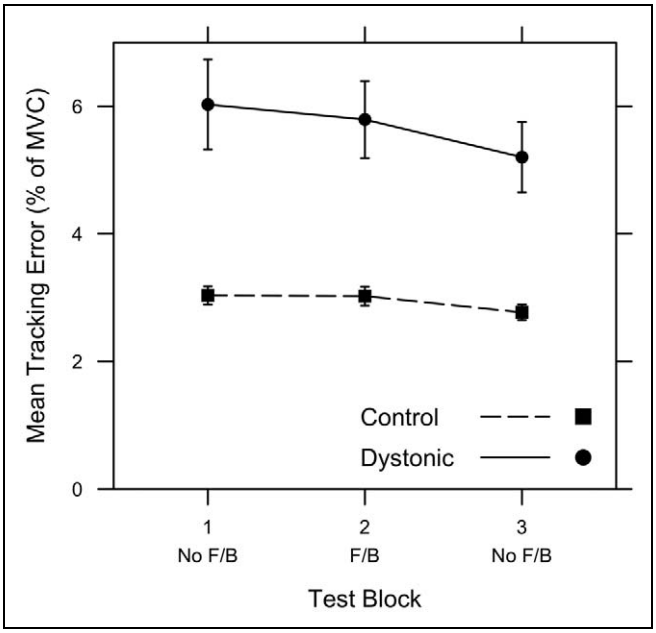

Figure 3. Mean tracking error for each group and block. Error bars indicate standard errors. MVC, maximum voluntary contraction; F/B, feedback.

= -0.341; $P = .733$), but it was significantly lower in Block 3 than in Block 1 ($t(404) = -3.27$; $P = .001$). Therefore, tracking error did not increase with decreased co-contraction.

We also tested whether the amount of co-contraction observed in the dystonic group was related to the severity of dystonia as measured with the Barry-Albright Dystonia Scale. To test this, we measured the Pearson correlation between the co-contraction values measured in Block 1 and the Barry-Albright Dystonia Scale scores from the tested arm. We found that co-contraction was positively correlated with the score on the Barry-Albright Dystonia Scale ($r = 0.678$; $P < .001$).

Discussion

To investigate the effect of visual feedback on co-contraction, children with and without dystonia performed an isometric tracking task that required alternate contraction of biceps and triceps muscles. We found that the dystonic group had greater levels of co-contraction than the control group, and the degree of co-contraction correlated with the severity of dystonia as measured with the Barry-Albright Dystonia Scale. When provided with visual feedback, individuals in both groups were able to reduce their levels of co-contraction, and they did so without increasing their error in the tracking task. When visual feedback was removed, both groups were able to maintain a level of co-contraction that was significantly lower than their initial levels, suggesting at least a short-term persistent beneficial effect of the feedback of co-contraction.

Most importantly, the results of this study suggest that children with dystonia can exert some control over their dystonic symptoms, if their attention can be drawn specifically to those symptoms. What might this role of augmented sensory feedback say about the nature of childhood dystonia? One possibility is that these children might not normally be aware of co-contraction. A theoretical study from our laboratory has suggested that the motor system could remain at a level of inadequate performance if it does not have access to sufficient sensory information.[23] Additionally, several studies have shown poor sensory performance in childhood dystonia, cerebral palsy, and adult dystonia.[24,25] Together with those previous results, this study supports the possibility that co-contraction and other symptoms of childhood dystonia might result from a lack of sufficient sensory feedback about motor actions, and these symptoms might be at least partially ameliorated by providing additional sensory feedback.

The prospect of using biofeedback as a therapy for childhood dystonia raises several research questions that were not considered in the present study. Most importantly, we have not investigated whether visual feedback can help individuals control co-contraction for any longer than a few minutes. The present study might be considered more similar to that of Lazarus and Todor,[12] in that we have identified that attention can play a role in the persistence of dystonic symptoms. Therefore, further studies are needed to evaluate the effectiveness of a longer-term therapy.[26] Additionally, future research will need to determine the effectiveness of a biofeedback therapy in terms of its effect on motor function and disability. Previous biofeedback approaches that have successfully modified muscle activity have not always had an effect on an individual's overall function.[3] Finally, it is also important to consider which individuals will respond to this form of therapy. Previous studies of biofeedback have often had variable results, with some participants not responding.[11,27] We saw evidence of a

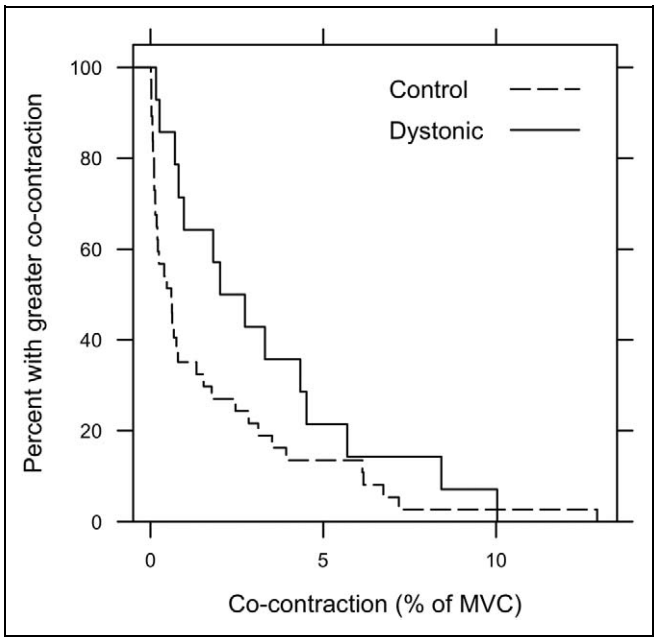

Figure 4. Kaplan-Meier plot showing mean co-contraction in Block 1 for individuals in the Control and Dystonic groups. Most participants in the Control group have co-contraction levels at or below 1% normalized electromyogram, and both groups have a small number of participants with co-contraction levels greater than 5% normalized electromyogram. MVC, maximum voluntary contraction.

similar variability in the present study, as some participants reduced their co-contraction much more than others.

If biofeedback training could help children gain an ability to control their symptoms, there are suggestions from related research that this might provide a lasting effect that goes beyond the focus of training. For example, Nash et al[11] found that learning the ability to control spasticity in 1 leg helped children to control the spasticity in their other leg. Dursun et al[10] showed that biofeedback training to help children reduce spasticity led to improvements in gait that continued to increase over the course of at least 3 months. Finally, a study in adult writers' cramp showed that biofeedback training was associated with changes in the density of dopamine receptors in the striatum, raising the possibility that this training can help bring about corrective changes in the neural pathways associated with dystonia.[28]

It is also important to point out that children with dystonia had greater co-contraction in this task than children without dystonia. We had expected this result, as co-contraction is considered to be an important symptom of dystonia,[15] and previous research has shown that individuals with dystonia have difficulty activating and relaxing muscles on demand.[29-31] A closer look at the results, however, provides a more nuanced picture. Figure 4 shows a Kaplan-Meier plot of the distribution of co-contraction in both groups during the first block of testing (without visual feedback of co-contraction). This figure shows that both groups had a similar range of co-contraction levels, from less than 1% normalized electromyogram to over 10% normalized electromyogram. One individual from the control group had more co-contraction than all participants in the dystonic group. Also, both groups had a skewed distribution of co-contraction values, with most participants having relatively low values, and a few with high values. This degree of skewness seemed to form the main difference between the groups, as the majority of children without dystonia had very low levels of co-contraction (ie, below 1% normalized electromyogram), whereas the majority of children with dystonia had moderate to higher levels of co-contraction. These observations indicate that the co-contraction levels seen in childhood dystonia are not completely outside the range of values seen in children without dystonia, but that moderate levels are much more common in children with dystonia than in those without.

The observed differences in co-contraction levels between children with and without dystonia raise questions about potential approaches for measuring dystonia. One of the most difficult aspects of studying dystonia in children is the lack of valid quantitative instrumented measures of severity, or a gold standard that ensures specificity of the measurements.[1] Several rating scales have been developed and tested, including the Burke-Fahn-Marsden scale[32] and the Unified Dystonia Rating Scale[33] for adults, and the Barry-Albright Dystonia Scale[17] and Hypertonia Assessment Tool[34] for children. In the absence of a gold standard definition, however, the specificity of these scales for the diagnosis of dystonia is unknown. Previous attempts at instrumented quantitative measures[35,36] have also shown promise, including correlation with clinical ratings of dystonia severity.[36] The work reported here suggests that quantitative measurement of co-contraction is possible, but the presence of co-contraction in control subjects can mean that this feature is not sufficiently specific to establish a diagnosis of dystonia. Although all of these approaches measure important aspects of dystonia, there is not yet a sufficient base of experience to be able to compare the methods. In the future, it will be very important to compare measures of co-contraction, overflow, and clinical scales to determine the relative sensitivity and specificity of these different measures and work toward the development of a true gold standard for the diagnosis and quantification of childhood dystonia.

Co-contraction between antagonist muscles is considered to be a hallmark of childhood dystonia. Using an isometric tracking task, we have shown that children with dystonia were able to reduce the level of co-contraction between their biceps and triceps muscles, provided that their attention was drawn to the co-contraction. These results help to shed light on possible underpinnings of childhood dystonia, as well as suggesting possible approaches for helping children to reduce dystonic symptoms in the future.

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Contributors

TDS conceived the experiment. TDS and JvD developed the apparatus and methods. JvD and SJY collected the data. SJY analyzed the data and wrote the first draft of the manuscript. SJY, TDS, and JvD revised the manuscript.

Declaration of Conflicting Interests

The authors declared no potential conflicts of interests with respect to the authorship and/or publication of this article.

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