Healthy and dystonic children compensate for changes in motor variability

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Chu VW, Sternad D, Sanger TD. Healthy and dystonic children compensate for changes in motor variability. J Neurophysiol 109: 2169–2178, 2013. First published January 23, 2013; doi:10.1152/jn.00908.2012.—Successful reaching requires that we plan movements to compensate for variability in motor output. Previous studies have shown that healthy adults optimally incorporate estimates of motor variability when planning a pointing task. Children with dystonia have increased variability compared with healthy children. It is not known whether they are able to compensate appropriately for the increased variability and whether this compensation leads to changes in reaching behavior. We examined healthy children and those with increased motor variability due to secondary dystonia. Using a simple virtual display, children performed a motor task where the variability of their movements was manipulated. Results showed that both subject groups changed their movement strategies in response to changes in the level of perceived motor variability. Both groups changed their strategy in a way that improved performance relative to the perceived motor variability. Importantly, dystonic children faced with decreased motor variability adapted their movement strategy to perform better and more similarly to healthy children. These findings show that both healthy and dystonic children are able to respond to changes in motor variability and alter their movement strategies.

A number of experiments on perception have shown that humans take uncertainty into account when interpreting and responding to sensory information (Bays and Wolpert 2007; Ernst and Banks 2002; Körding et al. 2004; Körding and Wolpert 2006). More recently, several studies on movement control have provided evidence that subjects choose movement strategies that minimize the cost of execution variability (Cohen and Sternad 2009; Harris and Wolpert 1998; Liu and Todorov 2007; Miyamoto et al. 2004; Müller and Sternad 2009; Scholz and Schöner 1999; Scholz et al. 2000; Sternad and Abe 2010; Sternad et al. 2011; Todorov 2004; Todorov and Jordan 2002; Trommershäuser et al. 2003). For example, Trommershäuser and colleagues showed that subjects modify their aiming movements to reduce the possibility of variability causing high-cost errors. Sternad and colleagues showed that subjects vary early in practice develop strategies that are tolerant to variability, even when such strategies do not minimize velocity or effort. These and other studies have provided evidence that humans compensate for their own motor variability when planning and executing motor actions.

Most previous studies have shown that humans take their own baseline variability into account when they adapt their strategy in response to changes in the pattern of reward and/or penalty (the cost function). However, most studies have not tested whether subjects respond appropriately when variability is manipulated experimentally. In only one study, by Trommershäuser et al. (2005), has it been demonstrated that healthy adults respond optimally to added display variability in a task where subjects reached to an end-point target near a penalty region. This shows that healthy adults incorporate observed variability in their movement planning. Compensation for artificially increased variability was seen immediately in a novel pointing direction, suggesting that subjects maintained an internal estimate of their task-relevant variability.

The ability to compensate for variability becomes particularly important when there are changes in intrinsic variability as, for instance, during development or fatigue, or in certain neurological disorders. In this study, we test children and how their responses change when their perceived variability is manipulated. The first goal of this study is to test the hypothesis that healthy children are able to compensate for changes in their own variability (hypothesis 1). We predict that healthy children will respond to increases in perceived variability by adopting a more conservative strategy that reduces the risk of failure.

The second goal of this study is to investigate whether children with increased variability due to secondary dystonia can adapt their behavior, despite their apparent diminished motor control. Dystonia in children is defined as a movement disorder in which involuntary sustained or intermittent muscle contractions cause twisting and repetitive movements, abnormal postures, or both (Sanger et al. 2003). When dystonia is the dominant manifestation of a genetic disorder, it is classified as primary dystonia. When dystonia occurs due to a recognized cause, such as trauma or stroke, it is classified as secondary dystonia (Tarsy and Simon 2006; Vitek 2002). Characteristic features of dystonia include movements that are slow (bradykinetic), inaccurate, and have high intrinsic variability (Chu and Sanger 2009; Malfait and Sanger 2007; Sanger 2006; Sanger et al. 2005).

We suggest that abnormal movement planning and speed in children with secondary dystonia may be in part a compensatory response to increased motor variability. We hypothesize that, like healthy subjects, children with secondary dystonia adapt their movement strategy when variability is manipulated (hypothesis 2). Therefore, we predict that when perceived variability is lowered in dystonic children, their movement strategy will change in a way that takes advantage of the perceived reduction in risk of failure, and the strategy may approach that of healthy children.

Our final hypothesis is that the changes in strategy in response to the variability manipulation occur in the direction of the optimal strategy for the new variability (hypothesis 3a). We therefore compare the mean strategy adopted by each child with the theoretically optimal one for that child in the specific task condition. We test whether these adaptations have similar magnitude in healthy children and those with dystonia (hypothesis 3b). If chil-
dren with dystonia can be made to perform more similar to healthy controls by manipulating their perceived variability, this provides evidence that the response to variability is a cause of the differences in motor performance.

MATERIALS AND METHOD

Participants

Sixteen children with secondary dystonia were recruited from the Lucile Packard Children’s Hospital at Stanford (ages 6–18 yr, 6 males and 10 females). All participants had a diagnosis of cerebral palsy (CP) with dystonia affecting one or both arms. Dysfunction and CP were determined by clinical examination according to standard definitions (Sanger et al. 2003, 2006). The experiment tested performance of the affected arm; when both arms had dystonia, the more affected arm was used. Subjects were excluded if there was spasticity, hyper-reflexia, or weakness in the tested arm. Sixteen individually age-matched control subjects (ages 5–18 yr, 12 males and 4 females) were recruited from the local area. Healthy subjects performed the experiments with their dominant arm. Details of the participants are provided in Table 1.

Healthy children were tested on their dominant arm, whereas dystonic children were tested on their more affected arm. Since one goal of the experiment (hypothesis 3) was to assess whether the healthy children perform more like dystonic children and the dystonic children perform more like healthy children, this strategy is the most conservative because it starts with the maximum possible difference in performance. If both groups show similar magnitude of change but opposite direction of change, this gives the strongest possible result. This strategy would not have been appropriate if the purpose were to compare performance between groups per se. The only comparison that causes a variable outcome \( x \) can be described by the conditional probability density \( p(x|u) \). Given a cost function \( V(x) \) that determines the cost of each possible value of the outcome \( x \), the goal of motor control is usually to find the command \( u \) that minimizes the expected value of the cost \( \mathbb{E}[V(x)|u] \). We thus have the following equation for the optimal motor command \( u^* \):

\[
u^* = \arg\min_u \mathbb{E}[V(x)|u] = \arg\min_u \int V(x) p(x|u) \, dx.
\]

On the basis of this model, if humans minimize the expected cost \( \mathbb{E}[V(x)|u] \) based on an estimate of the variability \( p(x|u) \), then we expect that the motor command \( u \) will change if either the cost \( V(x) \) or the estimated variability \( p(x|u) \) is altered. As mentioned above, previous studies have predominantly manipulated the cost function \( V(x) \); the present study manipulates \( p(x|u) \).

To test this hypothesis, the experiment used a shuffleboard task where children slid a virtual puck along a simulated table of constant friction to land as close to the edge of the table as possible without falling off. Points were awarded based on how close to the edge of the table the puck stopped. The number of points was made visually explicit to the subject using colored regions on the table surface. The final puck position was controlled by the velocity of the arm at the time the puck was released. Velocity at release was regarded as the control variable \( u \). The goal of the task was to maximize the total score in a given session, i.e., maximizing the expected value of the score. The optimal value of \( u \) depends on the noise distribution \( p(x|u) \). Since the purpose of this study was to measure the subject’s compensation for a change in motor variability, the experiment manipulated \( p(x|u) \). Increasing and decreasing variability in an experiment can be achieved in several ways. However, it is impossible

<table>
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<tr>
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<th>Sex</th>
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CP, cerebral palsy; M, male; F, female.

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to apply identical manipulations to achieve both increase and decrease. Hence, we reduced variability by smoothing over successive release velocities and increased variability by adding random Gaussian-distributed noise to the release velocity. The experiment tested changes in performance in dystonic children and age-matched healthy children. Given the high variability in dystonic children, their variability was decreased. In healthy subjects, variability was increased by a corresponding amount, as described below. The manipulations were designed to make the amount of change in both subject groups as similar as possible. Note, however, that the manipulations could not make the amount of variability identical in the two subject groups.

By comparing these conditions, we can evaluate how the children change their control $u$ in response to the changes in $p(x|u)$. If the healthy subjects change $u$ when $p(x|u)$ is increased, this would support hypothesis 1. If dystonic children modify their control $u$ according to decreases in $p(x|u)$, the results would support hypothesis 2. This result would demonstrate that dystonic children have the ability to flexibly adapt their movements to their variability, showing more motor control than often assumed.

In addition, we tested the direction and magnitude of change in the two subject groups and compared these values with the predicted optimal strategy (hypothesis 3a). For any level of variability there exists an optimal release velocity that maximizes the total score. It is important to emphasize that maximizing the total score given the presence of variability is different from maximizing the immediate single score without knowledge of variability. If subjects had only been given one trial and instructed to maximize a single score, they should have always aimed for the highest scoring region. Optimizing the total score over a series of throws instead requires taking variability into account. Referring to Eq. 1, this optimal strategy was $u^*$ and the cost function was $V(x)$.

Thus the optimal strategy corresponds to an optimal release velocity $u^*$ that maximizes the expected value of the total score $V(x)$ for a given standard deviation $\sigma$. If $x$ is drawn from a Gaussian distribution, with $u$ as mean and $\sigma$ as estimated variability, this is denoted as $N(u, \sigma)$.

$$u^*(\sigma) = \max \mathbb{E}[V(x) | x \sim N(u, \sigma)].$$

If the results show that compensation for variability is a significant cause of changes in target selection in dystonic children, then this renders new insights into the mechanism of motor abnormalities and could become the basis for new therapeutic interventions.

Experimental Apparatus

The experimental setup is shown in Fig. 1. Subjects sat in an adjustable chair. More severely affected dystonic subjects were permitted to remain in their own wheelchair. Elbow kinematics were measured using a single-joint manipulandum. The manipulandum was part of a motorized robot arm with the motor disabled. The subject’s forearm was strapped onto the manipulandum, which measured the angular position of the forearm using an optical position encoder on the motor (D063M-23-1310; Kollmorgen Direct Drive Rotary). The experimental setup restricted all forearm motion to the horizontal plane. The movement of the manipulandum was displayed on a screen in front of the subject.

A top-down view of a shuffleboard table was displayed on a computer monitor (Fig. 2). The movements of the manipulandum were shown by the white paddle at the bottom of the screen. Subjects were instructed to use a rapid extension movement of the forearm to slide a puck along the shuffleboard so that it stopped as close to the far edge as possible without going beyond the edge. The puck was restricted to move in a straight line along the table. The table showed five different color regions that represented areas with increasing scores: brown = 1, blue = 3, green = 6, yellow = 10, and red = 20. The score for each throw was determined by the region in which the puck stopped. If the puck fell off the far edge of the table, the trial scored 0 points. This created a reward function that monotonically increased, followed by a sharp cutoff (falling off the table). After each throw both the score of the current trial and the cumulative score of all previous trials were displayed on the monitor. The score regions corresponded to the following release velocities: brown, 0–57% $v_{max}$; blue, 57–63% $v_{max}$; green, 63–69% $v_{max}$; yellow, 69–75% $v_{max}$; and red, 75–80% $v_{max}$. Subjects who were unable to perform successful throws, i.e., land the puck on scoring regions of the table, in at least half of the trials were excluded from the study.

To initiate a trial, the subject flexed the elbow until the tip of the white paddle on the screen touched the lower orange line (Fig. 2). At this point the paddle “picked up” the puck. To throw, the subject extended the forearm. At the moment when the paddle crossed the upper orange line, the puck would leave the paddle, moving forward along the direction of the table. Its tangential velocity $u = vL$ was determined by the paddle length $L$ and angular velocity $v$ of the manipulandum at the time of puck release. The paddle length $L$ was set to be 0.4 m. As soon as the puck was released, it was subjected to a constant frictional force $mg\mu$, where $m = 0.1$ kg was the mass of the puck, $g$ was the acceleration due to gravity, and $\mu$ was the coefficient of sliding friction. The value of the friction coefficient was determined on the basis of each subject’s maximum release velocities measured before the experiment. The length of the virtual shuffleboard was set

![Fig. 1. Experimental setup. A: schematic showing the subject seated in a chair with arms strapped in to a manipulandum rotating around a single axis of rotation placed directly underneath the elbow. The manipulandum was used to measure the arm kinematics. PC: personal computer. B: arm manipulandum with the subject’s arm strapped in.](http://jn.physiology.org/)
100 baseline trials with no manipulations to the visual display. In block A, each subject group received their individually tailored manipulation of perceived variability.

For the dystonic subjects, the variability was decreased in block B; for the healthy subjects, variability was increased in block B. We did not test increased variability in dystonic children because their variability was already so high that increased variability would have made the task impossible for them. We did not test decreased variability in control subjects because the increased variability case was sufficient to test our hypotheses and additional test conditions would have unnecessarily prolonged the duration of the experiment. To facilitate comparisons between groups, we matched the increases and decreases in variability as described below.

For the dystonic participants, their displayed variability was reduced by modifying their actual velocity at release, \( v_{\text{actual}} \), to obtain a new velocity, \( v_{\text{display}} \), used to calculate the puck’s motion. For each trial \( i \), we calculated the moving average of the previous three trials and the current release velocity:

\[
v_{\text{display}, i} = \frac{1}{4} \sum_{k=0}^{3} v_{\text{actual}, i-k}.
\]

This was equivalent to temporal filtering and reduced the variance by a factor of 4 and the standard deviation by a factor of 2.

Subsequently, a variability reduction ratio \( r \) was quantified for each dystonic participant to serve as a basis for determining the increase in variability for his/her pairwise age-matched healthy subject. This paired manipulation ensured that the manipulated display variability for the healthy subjects was in the same range as the raw variability in the dystonic subjects. This ratio \( r \) was calculated as the standard deviation (SD) of the difference in release velocities, normalized by the display velocity:

\[
r = \frac{\text{SD}[v_{\text{actual}} - v_{\text{display}}]}{v_{\text{display}}},
\]

Using this reduction ratio \( r \), the actual velocity \( v_{\text{actual}} \) of healthy subjects was increased by an amount similar to the reduction in variability in their age-matched dystonic subject. Specifically, velocity-dependent noise scaled by \( r \) was added to \( v_{\text{actual}} \) of each healthy subject:

\[
v_{\text{display}} = v_{\text{actual}} + r \cdot v_{\text{actual}} \cdot \phi,
\]

where \( \phi \) was a Gaussian random variable with zero mean and unit variance. Velocity-dependent noise was chosen because previous studies have shown that variability in dystonic children is best described as added signal- or velocity-dependent noise (Sanger et al. 2005; Smits-Engelsman et al. 2007). Addition of noise rather than simple error amplification was chosen for healthy subjects because this was deemed to render a more appropriate structure of increased variability.

The manipulations for both the dystonic and healthy subjects were intended to keep the mean of \( v_{\text{display}} \) the same as the mean of \( v_{\text{actual}} \). However, due to the velocity-dependent noise, there was a slight bias since the tails of the noise distribution were longer for higher velocities.

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1Post hoc inspections of the added random noise \( \phi \) in the control subjects showed that there was a slight asymmetry in the distribution (a slightly longer positive tail). This was due to the small number of samples from the random number generator (\( n = 100 \)). The same random numbers were used for each subject. Whereas the median was unaffected (median = 1.25% \( v_{\text{actual}} \)), the mean had a bias that was compared with the added variability to assess its effect: bias = \( \sqrt{\text{Var}[v_{\text{display}} - v_{\text{actual}}]} / \text{Variance}(v_{\text{display}} - v_{\text{actual}}) \). The bias for all the subjects was 0.24. Since the bias was due to a longer tail in the same direction as the longer tail due to signal dependence of the noise, the additional bias would be perceived by subjects as a slightly greater magnitude of signal-dependent noise.
Position data of the manipulandum from the encoder were acquired by a data acquisition board (USB-1208F8; Measurement Computing) and stored on a personal computer using custom software written in Visual C++. Data analysis software was written in MATLAB (The MathWorks).

The main measure extracted from the continuous velocity recordings was the subjects’ single-sample velocity at release. The actual velocity $v_{\text{actual}}$ and, consequently, the displayed velocity $v_{\text{display}}$ were normalized to each individual’s maximum release velocity $v_{\text{max}}$ to afford comparison between subjects. Figure 3 illustrates the release velocities across the 200 trials in the two blocks in one healthy and one dystonic subject. The gray-shaded background signifies the score region where the puck would have landed given the respective release velocity. The red traces in block B show the increased and decreased velocity as displayed to the subject. Some oscillatory behavior was observed, especially in the dystonic subjects’ velocities in block B. This may have resulted from the real-time filtering that necessarily introduced a delay and potentially enhanced oscillatory behavior. However, these oscillations were small and unlikely to interfere with changes in the mean, which was the focus of analysis.

Three dystonic subjects could not complete all 200 trials. Their data were nevertheless included. A criterion for exclusion of the data was when a subject had more than 50% of failed trials in which they overshot the target and received zero score. One dystonic subject did have more than 50% failed trials and was subsequently excluded from the statistical analysis (more details are provided in RESULTS).

**Time series analysis.** To shed light on the subjects’ strategies in the shuffleboard task, we examined the time series of the release velocities. First, to identify trends in the release velocities due to learning or fatigue, we fitted the time series of 100 velocity estimates of each individual in each block by a linear regression. Such trends were considered insignificant if the slope was not significantly different from zero. Second, to test whether subjects used a simple compensatory strategy that reduced release velocity following any failed trial (the puck falling off the far edge; 0 score), we compared the velocity from trials immediately following a failed trial with the mean velocity across the last 50 throws of each block. If the subjects performed immediate corrections, we would expect that the mean of the velocities of trials immediately following a failed trial would be lower than the mean velocity of all trials. Third, to see if there were error-based adjustments to the release velocities, we examined the correlation between the displayed release velocity (from trial $i$) with the change in actual release velocity (from trial $i$ to $i + 1$) using data from the last 50 trials of each block. If low velocities were followed by consistent increases in release velocity, then an error correction adaptation strategy would be supported.

**Change in performance strategy: intended velocity and its variability.** The primary dependent measure was the actual velocity at release for each trial. For the statistical comparison of release velocities in blocks A and B, only the last 50 trials of each block were included where individuals approximated steady-state behavior (tests of stationarity are reported in RESULTS). Note that some dystonic children could not complete all 100 trials. We nevertheless used their last 50 trials, rather than half of the completed trials, because this made the statistical tests more conservative.

To obtain an estimate of the intended velocity $v_o$, we calculated means in windows of five trials; hence, the sequence of 50 trials was parsed into 10 nonoverlapping windows. For each window, the mean and its standard deviations were determined and used as dependent measures. To test hypotheses 1 and 2, means and standard deviations were submitted to a one-way (block) repeated-measures ANOVA; each group was tested separately. Hypothesis 3 was not evaluated for these two measures, because the velocity means and their changes due to increase and decrease in displayed velocity were not directly comparable. Statistical analysis was performed in SPSS, with the $α$ level set at 0.05.

**Deviations from optimality.** Hypothesis 3a stated that subjects compensated in an appropriate direction that would approach the optimal solution. To quantify this change in strategy $v_o$, we first determined the optimal velocity $v^*$ for a range of standard deviations. For each subject, we then used their estimates of the standard deviations $σ$ to calculate an optimal strategy $v^*$ for the last 10 five-trial windows. The deviation of $v_o$ from $v^*$ was computed in each five-trial window. Note
that in block A, \( v \) and \( \sigma \) were calculated from the actual mean and standard deviations of 10 throws; in block B, calculations of \( v \) and \( \sigma \) were based on the displayed velocities, \( v_{\text{display}} \). To test hypothesis \( 3b \), the two groups were compared in their adaptations. A 2 (subject group) \( \times \) 2 (block) ANOVA was conducted on this deviation measure. All analyses used an \( \alpha \) level of 0.05.

RESULTS

The range of maximum velocity \( v_{\text{max}} \) for the healthy subjects was between 7.81 and 15.44 rad/s, and that for the dystonic subjects was between 1.27 and 11.33 rad/s. Even though several dystonic subjects stayed considerably below the release velocities reached by healthy subjects, this did not affect the results since the shuffleboard length was normalized to their maximum velocity.

Each subject’s data were evaluated for their overall ability to perform the task. As a measure of success or failure, the number of trials where the puck failed to stay on the shuffleboard was determined. If the subject failed to keep the puck on the shuffleboard more than 50% of the trials, the subject was regarded as not having mastered the task. One dystonic subject (subject 15) had a failure rate of 62%. Hence, this particular subject did not meet inclusion criteria and was excluded from further analysis.

Figure 3 shows representative time series from a pair of subjects, one from each group. Given the normalization of the virtual shuffleboard, the velocity is shown as a percentage of maximum velocity, \( \%v_{\text{max}} \). It is apparent that both mean and variability were different across the two blocks in both subjects. When the displayed variability was increased for the healthy subject in block B, velocities were lowered, i.e., he chose a safer but less rewarding strategy than in block A (subject 1). In contrast, the dystonic subject performed with visibly higher variability in block A (subject 13). In block B, when variability was reduced, he adopted a strategy that presented similar risk but higher rewards.

A prerequisite for the subsequent comparisons of performance in the two experimental conditions was that there were no significant trends due to fatigue or learning. To this end, the time series of 100 velocity estimates of each individual in each block was fitted by a linear regression. For the dystonic subjects, none of the regression slopes were statistically different from zero. For 3 of 16 healthy subjects, the regression slopes showed a significant positive slope in block A (subject 1). In contrast, the dystonic subject 7 had a rightward bias in the dystonic subjects. The histograms also show how the distributions of the actual velocities in block B shifted, shown in red outline. As hypothesized, the healthy subjects shifted the peak of their release velocities to lower values; in contrast, the dystonic subjects biased their release velocities to higher values. The pink lines represent the displayed or manipulated velocities.

The changes in intended release velocity (mean across 5 trials) across blocks were evaluated for both subject groups separately, using one-way repeated-measures ANOVAs. For the healthy chil-

![Healthy Subjects and Dystonic Subjects](http://jn.physiology.org/)

**Fig. 4.** Distribution of release velocities from the dystonic and healthy subjects in the 2 experimental conditions. The peak of the distribution shifted to the left in the healthy subjects and had a rightward bias in the dystonic subjects. \( v_{\text{actual}} \): actual velocity at release; \( v_{\text{display}} \): displayed velocity.
The velocity means decreased significantly from 71.61%\(v_{\text{max}}\) in block A to 66.21%\(v_{\text{max}}\) in block B [\(F(1, 144) = 96.44, P < 0.0001\)]. Similarly, the dystonic children increased their mean velocity from 67.65 to 70.21%\(v_{\text{max}}\) [\(F(1, 131) = 10.63, P < 0.001\)]. The two results are in support of hypotheses 1 and 2. Figure 5 shows the means of all subjects in blocks A and B. The subjects are rank-ordered by their mean velocity in block A. As shown, all 16 healthy subjects lowered their mean release velocity from block A to block B. In the dystonic group, 10 of the 15 dystonic subjects increased their mean release velocity from block A to block B. The amount of change tended to be higher when the initial release velocity was lower. Four of the five dystonic subjects who did not increase their mean release velocity had the highest mean release velocities in block A. This indicates that they may already have performed close to their maximum velocity and could not increase their velocity further. Ordering the subject data by age did not reveal any pattern.

It is possible that subjects had some ability to control their intrinsic variability and could thereby compensate for our intervention. To test this, the standard deviations of the actual velocities in the 10 windows were assessed for changes from block A to block B, using one-way ANOVA for each group. Neither of the two groups showed a significant change in their intrinsic variability across the experimental conditions. For the healthy subjects the standard deviations were 8.19%\(v_{\text{max}}\) in block A and 8.42%\(v_{\text{max}}\) in block B; for the dystonic subjects they were 13.69%\(v_{\text{max}}\) in block A and 12.30%\(v_{\text{max}}\) in block B. Thus the effect of manipulations on the intrinsic motor variability was negligible. This lack of change in variability is important for the subsequent calculations of the optimal \(v^*\), because they rest on the assumption that subjects’ intrinsic variability remained constant throughout the experiment.

Figure 6 shows the calculated optimal release velocity \(v^*\) together with subjects’ actual intended release velocity \(v_o\). The optimal velocity \(v^*\) was calculated for a range of standard deviations \(\sigma\) and is indicated by a solid line, which is the same line in all four panels. As expected, with increasing \(\sigma\) the optimal velocity \(v^*\) declines. The horizontal dashed line indicates the 75%\(v_{\text{optimal}}\), which is the minimum release velocity that would allow the maximum possible score for any single throw. Each data point represents the average intended release velocity \(v_o\) of each subject plotted against its measured standard deviations \(\sigma\) (averaged over the last 10 five-trial windows). The two orthogonal error bars show the standard errors in \(v_o\) and in \(\sigma\). Note that the velocity used in the calculations for block B was the displayed velocity.

The first test of hypothesis 3 evaluated whether subjects’ actual performance was different from the optimal strategy. We conducted \(t\)-tests for each individual subject comparing the difference from optimality against zero. In the healthy group, 5 of 16 individuals differed from optimal in both blocks; in the dystonic group, 6 and 7 subjects differed from the optimal strategy in blocks A and B, respectively. (Bonferroni corrections were not applied, because they would bias the tests toward the hypothesized result.) Sign tests based on the binomial distribution showed that the probabilities of being non-optimal in the four groups were 0.11, 0.11, 0.50, and 0.30. On the basis of this very course-grained test, it can be concluded that most of the children in both groups performed close to the optimal solution.

A more fine-grained test of hypothesis 3 compared the two subject groups in their adaptations from block A to block B. Figure 6 highlights the low-variability condition (block A in healthy, block B in dystonic) and the high-variability condition (block B in healthy, block A in dystonic). From Fig. 6, we see that healthy subjects deviated more from the predicted line in block B compared with block A, whereas dystonic subjects clustered around the predicted line similarly to the healthy subjects when their variability was lowered. Two dystonic subjects (subjects 7 and 8, marked by black circles) significantly undershot the optimal solution \(v^*\) in both experimental conditions by more than three standard deviations from the rest. These two subjects were also unable to finish the trials (only one other subject terminated the experiment prematurely) and were excluded in the following analysis. To statistically compare the deviations from the optimal strategy between groups and blocks, the difference between \(v_o\) and \(v^*\) for each subject was subjected to a 2 (subject group) \(\times\) 2 (block) repeated-measures ANOVA. The interaction was weakly significant [\(F(1, 304) = 4.05, P < 0.045\)], showing a small decreasing trend in optimality in controls and a small increase in dystonics. No other effect was significant. Aside from the
two dystonic children who had significant deviations from the optimal solution, the absence of a main effect for group signified that healthy and dystonic children did not statistically differ in their degree of optimality. Note that optimality in this study was defined specifically to the release velocity in the context of the shuffleboard task.

**DISCUSSION**

The study tested whether healthy and dystonic children are able to adapt to changes in perceived motor variability. We further tested this ability in two groups for whom adaptation to change in variability has particular relevance: children (a period of motor development) and children with increased variability due to dystonia. These are the first experiments to test the interaction between variability and task constraints in children and the first experiments to test such responses in children with dystonia.

**Hypothesis 1**

The first hypothesis in this study was that children readily adapt to changes in movement uncertainty, even at an early age. Our results showed that children from age 5 to 18 yr modified their movement strategies in response to increases in the perceived variability. The results showed that increasing variability in the healthy children caused them to take a more cautious strategy. In the present shuffleboard task they aimed toward score regions with lower release velocities, farther away from the table edge. Hence, these results support the hypothesis that children are able to change their motor plans rapidly and appropriately in response to ongoing changes in variability.

**Hypothesis 2**

The second hypothesis was that dystonic children, who have highly variable movements, are also able to adapt to changes in...
variability. The results demonstrate that despite their often seemingly uncontrolled movements, children with dystonia can respond to changes in motor variability. This flexibility was observed in 10 of 15 children, including the youngest subject. Children increased their release velocity and threw “harder.” Given that the task was not easy for most of the children, and some could not even complete the entire set of trials, to observe a significant increase in velocity is even more remarkable. As Fig. 5 suggests, individuals who did not take the opportunity to throw further when variability was lowered were those who already performed with high velocities, i.e., with high scores. It is also noteworthy that this phenomenon was already seen in the youngest among the participating healthy children, at the age of 5 yr. When the changes in velocity were examined for age effects, no systematic trends were visible.

Hypothesis 3

The third hypothesis was that all individuals, regardless of whether dystonic or healthy, move toward an optimal strategy for their particular level of variability. Individual pairwise comparisons with the optimal strategy showed that the majority of subjects in both groups did not differ from the optimal strategy. Although the relative number of nonoptimal individuals was slightly higher in the dystonic group, a binomial test did not render this difference as significant. When the deviation from optimality was directly compared across the two subject groups, healthy subjects did not show more optimality than dystonic children. This suggests that dystonic and healthy children may have a similar ability to compensate for variability. Furthermore, since the compensations were in opposite directions (healthy children approached the baseline performance of dystonic children, and dystonic children approached the baseline performance of healthy children), this result is consistent with the hypothesis that a significant component of the difference in aiming behavior (different average release velocities at baseline) between the two groups is explained by compensation for differences in intrinsic variability.

Compared with most studies in adults, these results were obtained in relatively short experimental sessions with only 200 trials (cf. ~1,200 trials in adults in Cohen and Sternad 2009, 2012). This does not allow much familiarization with experimental conditions. The practical reason is that it is difficult and oftentimes not possible to obtain long series of trials from children, particularly those with dystonia. Hence, the fact that significant results were obtained is all the more surprising.

Handedness

Are the results in dystonic subjects confounded by different hand dominance? As noted, dystonic subjects performed with their affected hand, which was the right hand in 7 of 15 subjects (subject 15 was excluded). If hand dominance had an effect on task performance, the expectation would be that the dystonic subjects, who were performing the task with their nondominant arm, would perform worse. Assuming a high likelihood of right-hand dominance, the expectation that dystonic subjects with right-hand performance scored better was not supported: dystonic subjects who were tested with their left arm had even better baseline performance than those who were tested with their right arm: mean release velocity, 70.49%v_{max} (left arm) and 63.07%v_{max} (right arm) (P < 0.0001). That aside, as mentioned above, for children who have sustained an early insult to the brain there is no meaningful way to assess hand dominance. More importantly, this experiment does not compare healthy and dystonic performance per se. Rather, we test whether dystonic children can modify their strategy in similar ways as healthy children when their variability is modified. Hence, to compare the performance of the affected arm in dystonics with the dominant arm in healthy children presents a very conservative test.

Our research is an extension of a number of studies that examined learning and adaptation of movement strategies. Previous work suggested that task learning involves exploring the cost landscape, and planning of motor commands is based on the expected cost (Sternad et al. 2011; Todorov 2004; Todorov and Jordan 2002; Trommershäuser et al. 2003, 2005). In this study, we have examined behavior in a group of subjects that have increased intrinsic variability. Similar to a study by Trommershäuser et al. (2005) in adults, our results are consistent with the hypothesis that children maintain an internal estimate of their own variability. This is also consistent with the speed-accuracy trade-off in Fitts’s law, where planning of movement speed depends on the available target width, i.e., permitted variability. The novel aspect of this study is that we focused on the estimation of variability in velocity rather than in position. Estimating variability in release velocity has an added layer of complexity because it cannot be directly observed or modified using feedback during movement. Furthermore, we tested the response to reduced variability (condition B in the dystonic subjects) and showed that dystonic children demonstrate the predicted compensated behavioral strategy. Although our findings show changes in strategy consistent with an internal estimate of variability, our results cannot conclusively rule out a different mechanism in which a change in the release velocity is based on observation of the immediately preceding error. Because the variability is equally likely to cause overshoot as undershoot, this would require a recursive adaptation algorithm that takes the cost function (the shuffleboard scoring structure) into account so that the magnitude and direction of response to error appropriately adjust for the effect on cost. If, for example, the shuffleboard were reversed, the appropriate response to variability would need to be reversed so that increased variability would require throwing farther. Our trial-by-trial analysis suggests that a single-trial adaptation rule is unlikely to explain the data, but future experiments are needed to determine which strategy is used by children to adapt to changes in variability.

Note that our approach depended on the fact that the shuffleboard task had an asymmetric cost function with a discontinuity. Real life examples with similar asymmetry include carrying a cup of coffee without spilling or hiking on a path that is near a cliff. If the task had a symmetric cost function, changes in the magnitude of variability would have no effect on the optimal strategy. The present results are consistent with recent findings showing that subjects choose strategies that tolerate variability, even if these strategies are close to a discontinuity in the cost (Hasson et al. 2012; Sternad et al. 2011). It is interesting to examine inter-individual differences in risk attitude. Nagengast et al. (2009) showed that individuals trade off between mean performance and variance when choosing a movement strategy, albeit in different degrees.
We do not know whether subjects interpret changes in variability as changes in perception or changes in motor function. The latter seems more likely, because the display did not change. Since we examine changes in strategy in response to changes in perceived variability, our results and interpretation are not affected by how the subjects interpret the change in variability. Nevertheless, this would be an important future research question and might be particularly interesting in subjects with sensory deficits that could increase sensory uncertainty.

Conclusion

Our results show that children compensate for perceived movement variability when performing motor actions, even at an early age and even in neurologically impaired children with dystonia. Healthy children and those with dystonia approach more optimal strategies given their specific variability, and there was no qualitative difference in the ability to compensate between groups. The results have important implications for understanding movement control in dystonic children. It is well documented that movement in dystonic children is both slow and variable (Chu and Sanger 2009; Sanger 2006; Sanger et al. 2005). We conjecture that movements are slowed partially due to compensation for variability. These observations suggest that children with dystonia are able to infer appropriate strategies that compensate for their movement disorder, and our results could therefore be helpful in guiding the design of new therapies.

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DISCLOSURES

No conflicts of interest, financial or otherwise, are declared by the authors.

AUTHOR CONTRIBUTIONS

V.W.T.C., D.S., and T.D.S. conception and design of research; V.W.T.C. data collection; V.W.T.C. and D.S. performed experiments; V.W.T.C. and T.D.S. analyzed raw data; V.W.T.C. and D.S. interpreted results of experiments; V.W.T.C. and D.S. prepared figures; V.W.T.C. and D.S. drafted manuscript; T.D.S. edited and revised manuscript; V.W.T.C., D.S., and T.D.S. approved final version of manuscript.

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