Random perturbation: A potential aid in treatment of children with cerebral palsy

S. Bar-Haim [ab]; N. Harries [b]; M. Belokopytov [b]; E. Lahat [b]; J. Kaplanski [a]

[a] Faculty of Health Sciences, Ben-Gurion University of the Negev, Beer-Sheva [b] Motion Analysis Laboratory, Assaf-Harofeh Medical Center, Zerifin, Israel

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S. BAR-HAIM[1,2], N. HARRIES[2], M. BELOKOPYTOV[2], E. LAHAT[2] & J. KAPLANSKI[1]

[1]*Faculty of Health Sciences, Ben-Gurion University of the Negev, Beer-Sheva, and* [2]*Motion Analysis Laboratory, Assaf-Harofeh Medical Center, Zerifin, Israel*

Abstract

Background and purpose. The motor behaviour of children with cerebral palsy (CP) can be viewed in terms of a stable mode with very little flexibility that prevents adaptation to tasks. We hypothesized that the use of random perturbations (RP) would weaken excessive stability, introduce flexibility and enhance the effects of physical treatment. The objective was to evaluate the contribution of RP to gross motor function and mechanical efficiency (MEg) during intensive physiotherapy in children with CP.
Methods. A convenience sample of 20 children with CP (mean age 8.2, range: 5.9–12.9 yrs) were matched by age and GMFCS level, and randomly assigned to structured intensive treatment (SIT) or to SIT + RP groups. Groups received one month of daily treatment. RP was applied by engine-induced random passive cycling for upper and lower limbs for up to 10 min in a 90-min treatment session. Gross Motor Function Measure (GMFM)-66 and gross mechanical efficiency (MEg) during stair climbing (MEg) were measured before and after treatment.
Results. GMFM-66 scores increased by about 1.0 in both groups. However, external work and MEg increased significantly more in SIT + RP than SIT. The increase in MEg in SIT + RP was independent of the level of motor function at baseline.
Conclusion. The addition of RP in treatment of children with CP may have weakened previously established stereotypical motor patterns and introduced flexibility, thereby improving mechanical efficiency of a complex motor task. RP may enhance the effects of intensive treatment.

Keywords: *Cerebral palsy, dynamic system, mechanical efficiency, physical treatment, random perturbation*

Introduction

This study incorporates a 'dynamic systems' approach in conceptualizing the control of voluntary movements. Stability is an important movement characteristic within this approach. Stability of movements may be viewed as a positive feature that allows motor goals to be achieved under conditions of unexpected changes in environment. However, high stability also acts against adaptive changes in movement patterns and can be viewed as preventing movement flexibility and optimization.

Organization of motor functions in typical and atypical motor behaviours is a dynamic process. Accordingly, the individual and environment can be considered as one complex dynamic system, connected interactively by afferent pathways conveying sensory information about the environment to the individual, and efferent pathways effecting changes within the environment.

Under the regime of these multiple interconnections, the system tends to attain stable states that are interpreted as preferred behavioural states. Optimal energy efficiency and environmental constraints are included in this theory [1,2]. Optimal mechanical efficiency characterizes skilled motor performance, quantifying the ability to complete the task with minimum energy expenditure [3–5].

Dynamic systems are systems that change over time and the representation of a dynamic system requires the description of behaviour states in one or more dimensions. The descriptions of instantaneous changes in time can be characterized as the 'trajectory' of the dynamic system. This trajectory

(i.e. the evolving state of the system) can be mapped in phase space [6].

We can learn about the state of a complete system of properties by observing kinematic variables such as angular velocities. These variables are used to quantify the stability of the system and thus characterize it.

According to the International Workshop on the Definition and Classification of Cerebral Palsy, 'Cerebral palsy (CP) describes a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain' [7, p. 572].

In children with CP, early brain damage results in the channeling of motor development into pathologically restricted patterns. Limited repertoires of motor behaviours limit the feedback that these children receive from the environment, reinforcing the secondary peripheral pathologies (of muscles and bones) and curtail the development of new motor patterns. The motor behaviour of children with CP is an optimal outcome, based on individual constraints such as spasticity, high level of co-activation of agonist and antagonist muscles at a joint, and high energy cost [8]. Therefore, some of this motor behaviour can be described as 'hard-wired,' 'stereotyped' or 'obligatory' [9]. Stereotypy and resistance to change characterize these atypical movement patterns.

These concepts may be illustrated by examples, employing the motor task of cycling on a stationary bicycle, where the child was asked to pedal at a self-paced speed for one minute as smoothly as possible. Cycling movements are used to identify motor control mechanisms necessary to accomplish specific motor tasks [10]. The degrees of freedom in cycling are reduced because the motion of the feet is constrained to follow the crank path, making the angular velocities and accelerations the only variables. Angular velocity measurements (in radians/sec) are obtained at precise time intervals from the electronically tracked pedal movements. For the purpose of defining stereotypy and changes in behaviour, recurrence plots are made of these angular velocities [11]. Examples are given in Figures 1 and 2 of the utility of these plots. Figure 1A shows a typical recurrence plot of a younger (3-year-old) and Figure 1B of an older (10-year-old) normally developed (ND) child. The difference between the means indicates the difference in work done, and the difference between standard deviations quantifies the greater stability of the motor behaviour in the older ND child compared with the younger child, i.e., 5.9 ± 2.3 rad/sec in A and 9.3 ± 0.7 rad/sec in B. Figure 2 shows the recurrence plot of a 9-year-old child with CP,
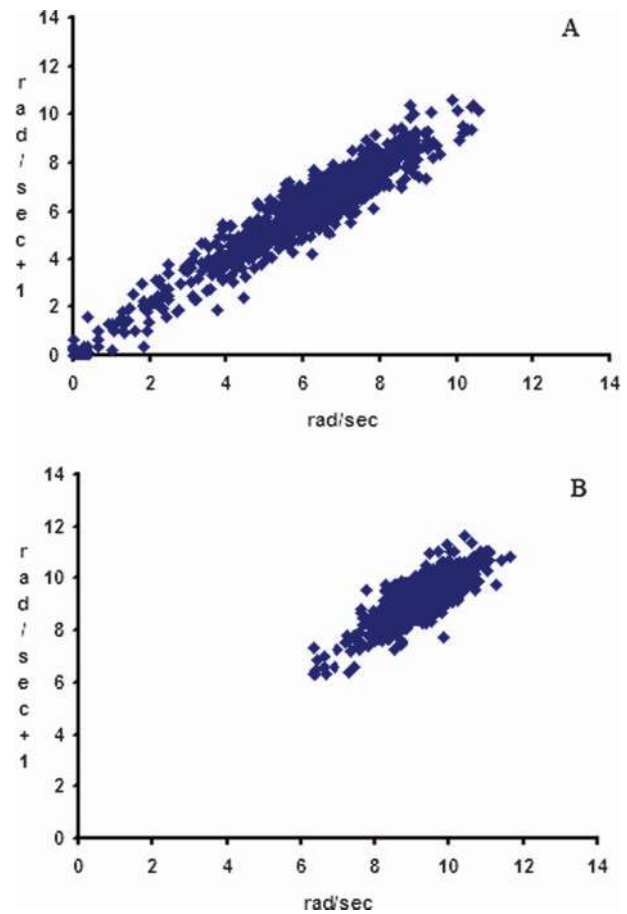

Figure 1. Top (A) shows a typical recurrence plot obtained during self-paced cycling (mean $\pm$ standard deviation of rad/sec) of a younger (3-yr-old) child (5.9 ± 2.3), and below (B), of an older (10-yr-old) normally developed (ND) child (9.3 ± 0.7). The means are directly related to the work done and the standard deviations are inversely related to the stability of the motor behaviour. In these plots the velocity in the first time interval (0.06 sec) is x_1, the velocity in the second is x_2 and y_1, the velocity in the third is x_3 and y_2, etc.

classified as level III according to GMFCS, before (Figure 2A) and after (Figure 2B) therapy. Before intervention the self-paced cycling velocities are almost zero rad/sec, indicating strong stability. Following therapy, the cycling velocity increased with increased variability, indicating more flexibility of the motor behaviour, i.e., 0.1 ± 1.3 rad/sec in A and 3.6 ± 3.3 rad/sec in B.

In terms of a dynamic systems approach, a change in behaviour can be induced by different types of external perturbations, thus potentially opening the door to new intervention strategies. For example, attempts are being made to control epileptic brain seizures by waiting for the system to approach an unstable point from the stable base. It then takes a minimal intervention to bring the system back to stability [12]. In another application, controlling cardiac arrhythmia, Garfinkel et al. [13] developed a method called 'proportional perturbation feedback,'
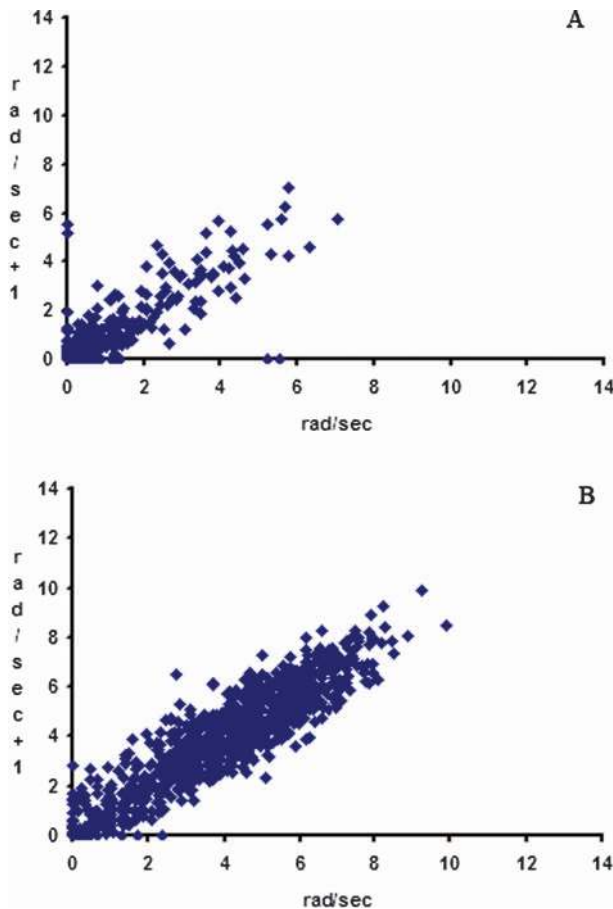
Figure 2. Top (A) shows the recurrence plot of a 9-yr-old child with CP, before therapy, and below (B), after therapy at a later date. A indicates a 'stiff attractor,' as cycling velocities are 'attracted' to near zero $(0.1 \pm 1.3$ rad/sec). In B, the mean velocity increased with increased variability, indicating more flexibility $(3.6 \pm 3.3$ rad/sec). See legend to Figure 1.

which was considered to perturb the system state point to move it toward the stable manifold. Other studies have demonstrated that applying low-level random vibrations to the feet can significantly reduce the postural sway during quiet standing in elderly subjects [14,15]. Shumway-Cook et al. [16] demonstrated that postural balance recovery and stability was improved following training by mechanical perturbations in children with CP. Woollacott et al. [17] suggested that reactive balance training by mechanical perturbations could result in changes in specific neural factors of balance control.

In the present study a unique intervention program was used that employs random movement perturbations (RP) to attempt to loosen highly stable motor behaviours and facilitate motor flexibility and learning. We hypothesized that RP would take the system out of its stable state and facilitate improved motor functions. We reasoned that RP induced by an external force moving limbs passively and randomly, requiring 'active relaxation' by the motor controller

[18], would prevent the child from relying on stereotypical habits, thereby increasing system flexibility. The incorporation of RP within an intensive physical treatment program could serve to improve the outcomes for children with CP. The primary objective of this study was to evaluate the contribution of RP to the improvement of gross motor functions and mechanical efficiency in children with CP during an intensive course of physiotherapy.

Methods and procedures

Participants

The local Institutional Review Board approved the study. The study was advertised and parents of children with CP responded. Written consent to participate was obtained from parents of children with CP who responded after they and the children were given a detailed explanation of the study. A pediatric neurologist screened the patients' medical history and a pediatric orthopedic surgeon determined the children's orthopedic status before they were asked to participate. During their first visit to determine study eligibility, a pediatric physiotherapist with 25 years experience with children with CP, classified the children according to the GMFCS for CP [19].

All of the children were taking part in continuous physical therapy programs of varying intensities in their local community or school. The inclusion criteria were: (a) diagnosis of CP, (b) age: 6–12 yrs, (c) level II, III or IV by GMFCS rating, (d) no orthopedic surgery or spasticity-reduction intervention during the previous 6 months, and (e) no major contractures of lower limbs muscles.

Twenty children, 14 boys and 6 girls, met the requirements and constituted the convenience sample. Their mean age was 8.2 yrs (range: 5.9–12.9 yrs). Participants were matched by age $(\pm 1$ yrs) and GMFCS level, and assigned to one of two study groups: the control group, whose physical therapy was a structured intensive treatment (SIT) and the study group who received SIT with the addition of RP (SIT + RP). The gender, age, anthropometric measures, type of CP and limb distribution by group are shown in Table I. There were no significant differences in age and anthropometric measures between the two groups at the beginning of the study.

Protocol

Both groups were tested at baseline and within 3–5 days after completing one month of SIT or SIT + RP therapy. Both groups received daily treatment sessions for 1.5 h, five days per week for four weeks (20 sessions).

Table I. Means ($\pm$SD) of age, physical characteristics, GMFCS classification and CP types for 2 study groups.

Characteristics	SIT ($n = 10$)	SIT + RP ($n = 10$)
Gender (male/female)	7/3	7/3
Age (years)	7.9 (1.6)	8.6 (1.9)
Height (cm)	118.5 (9.5)	121.2 (14.4)
Weight (kg)	21.9 (7.8)	25.7 (8.9)
Rt. Leg length (cm)	61.6 (6.1)	61.4 (12.0)
GMFCS II	2	2
GMFCS III	5	6
GMFCS IV	3	2
Diplegia – spastic	5	5
Diplegia – ataxic	1	–
Triplegia – spastic	1	–
Quadriplegia – spastic	–	3
Quadriplegia – mixed	3	2

Interventions

The same physical therapist treated all the children in the two groups and the treatment was carried out in the same environment for all participants. The study required 4 months to complete.

The SIT treatment consisted of an intensive, well structured, treatment protocol that included 7 – 10 min of passive stretching through a full range of motion of all limb muscles and an individual program of exercises of functional weight-bearing gross motor activities related primarily to locomotion. Each session included walking activities suited to individual abilities, e.g. playing with a ball while standing, jumping on a trampoline and climbing. Because of the individual limitations of each child in performing the ambulation skills, the training could not be standardized. However, each child was trained in at least three locomotor activities that included standing up from sitting, walking on different terrains and climbing stairs. Children classified as level II and III, according the GMFCS, ran, jumped on trampolines and climbed ladders. Children were made to walk on different surfaces at different speeds, with assistance and support given when necessary.

RP was applied by engine-induced passive cycling. An active passive trainer (APT-Tzora Active Systems, Kibbutz Tzora, Israel[1]) was modified with four separate motors and an electronic control board to randomize the rotation of each motor by speed, duration and direction (Figure 3). This allowed the engines to induce random passive rotary movements for each of the upper and lower limbs. The speed randomization ranged from 0 – 12 rad/sec and duration from 0.5 – 15 sec. The randomization allowed unpredictable changes of speed and direction (forward and backward) for each limb, for up to four limbs simultaneously (Figure 4).

Before RP interventions, the 10 children in this group were allowed to manipulate the modified APT

for familiarization before adjusting it for each child's height, leg and arm length. The children wore regular shoes without orthoses. Feet were inserted into special pedals and strapped with Velcro fasteners and the children grasped the handgrips, with special gloves if needed. They were asked to relax and allow the modified APT to move their limbs, without resisting.

RP was applied at the beginning of each session, for 2 min at the first, increasing to 10 min by the end of the first week. These 10 min were included in the 1.5 h treatment.

Preliminary trials indicated that the application of RP to the 4 limbs simultaneously was not well tolerated and therefore a sequence was chosen where, for 2/3 of the time that RP was applied, it was applied to both legs and for 1/3 of the time to both arms. The majority of the children enjoyed the RP treatments after the initial adaptation to the unpredictability. Following RP they continued with their regular SIT treatment session.

Tests and measurement

The Gross Motor Function Measure (GMFM)-66 tool was used to evaluate and compare the basic motor skills before and after one month of treatment. This is an ordinal measure designed to evaluate changes in gross motor function in children with CP in five motor dimensions (88 total items): (a) Lying and rolling, (b) crawling and kneeling, (c) sitting, (d) standing and (e) walk-jump-run activities. Specific items are scored on a 4-point ordinal scale [20]. The scores of these items were subsequently analysed using the gross motor ability estimator (GMAE) computer-scoring program to obtain a GMFM-66 score [21]. All tests were performed in the same room by a certified physiotherapist [22], who was not involved in the therapy and remained uninformed of the treatment groups to which each child belonged.

The gross mechanical efficiency (MEg) of a stair-climbing task was chosen as an ambulatory function measurement to compare the overall efficacy of interventions. MEg is a measure of the external work, divided by the total energy expended to produce it. The efficiency of human locomotion in walking, running and cycling ranges from 20 – 30% [23]. The MEg for ND children in stair-climbing is approximately 20% and that for children with CP is typically < 5% [24].

MEg was calculated for a $\sim$4-min climbing period as: External work (W)/total oxygen consumption (W). Oxygen consumption was measured with a portable K4b^2 analyser (COSMED, model Srl, Rome, Italy). The dynamic stair trainer (DPE Medical, Shoeva, Israel) was used for measuring external work. It has 5 stairs, adjustable
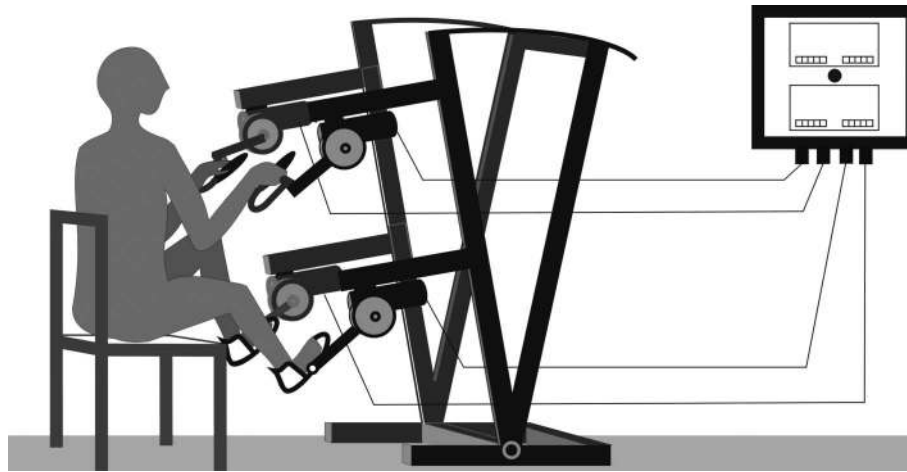
Figure 3. A schematic representation of the modified APT with four separate motors and an electronic control board to randomize the rotation of each motor by speed, time and direction. During the random stimuli (RP) intervention the children were seated and relaxed, allowing the APT to move their limbs passively.
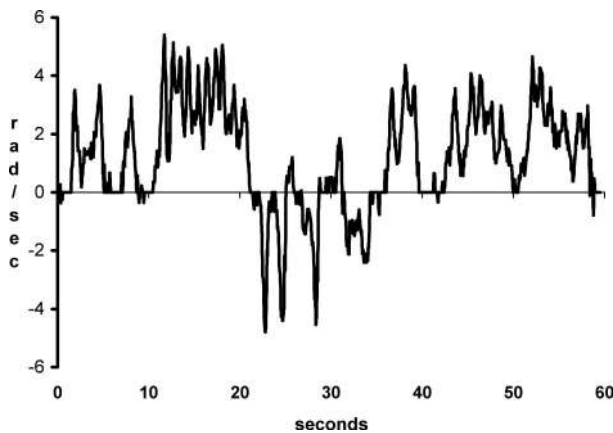

Figure 4. Velocities (rad/sec) recorded for one minute from a marker placed on the pedal of a lower limb during application of RP applied by engine.

from 0 – 17 cm, with adjustable handrails. The stair height was set according to each child's climbing ability during pre-test trials.

The distance component of the work over a finite time period is calculated from the summed vertical displacement of the body (m). The force applied is the body weight (kg) and the work is calculated as the product (kg × m), i.e., external work = body weight × number of stair ascents × stair height. The children climbed stairs at a self-paced speed because at that speed their performance is optimal and MEg is highest [25].

Each child was given a detailed explanation of the equipment and practiced stair climbing. One trial served as a practice test for familiarization. When the child felt comfortable, baseline measurements were made while sitting on a chair with backrest for 5 min and being told a story. Children then walked up and down the stairs continuously for ~4 min at a

self-paced speed, using the handrails for assistance if desired. After ascending to the top step they turned and descended in the opposite direction, the number of ascents being counted. Children with level IV GMFM were assisted with locating their hands on the handrails, but not with lifting their body to the next stair.

Statistical analysis

Data are presented as means (± 1.0 SEM). Baseline differences in measurement values between groups were tested for significance ($p = 0.05$) by two-sample t-tests assuming unequal variances. Differences in changes between the two groups from baseline to one month after intervention were similarly tested and changes within each group from baseline to one month were tested by paired t-tests. Least squares linear regressions were used to obtain significance of correlations between baseline values and changes from baseline for GMFM-66 and external work, O_2 consumption and MEg. An F-test was used to test the significance of difference in slopes of linear regressions.

Results

GMFM-66 and stair-climbing measurements of both groups of subjects before and after therapy are shown in Table II.

GMFM-66

For SIT, the baseline scores ranged from 39 – 85. The small average increase of 0.9 after treatment was not significant ($p = 0.06$). For SIT + RP, the baseline score ranged from 37 – 75, with the mean increase of

Table II. GMFM-66 and stair-climbing measurements of both groups before and after therapy.

Therapy	GMFM-66 Scores		Ext. work (W)		O₂ consumption (W)		Efficiency (%)	
	Before	After	Before	After	Before	After	Before	After
SIT								
Mean	56.7	57.6	3.03	4.23[#]	101	142	3.21	2.95
SEM	4.3	4.5	0.86	1.16	20	30	0.82	04.3
SIT + RP								
Mean	55.5	56.6[#]	2.70	5.56	156&	193	1.67	2.38[#]
SEM	3.3	3.2	0.86	2.27	10	25	0.47	0.51

W: work units (Watts); [#]value significantly greater than before and value significantly greater than SIT.

1.1 being significant. The increase in scores in both groups was not significantly different.

Gross mechanical efficiency (MEg) of stair climbing

The mean external work in SIT increased significantly after treatment by 1.2 W ($p = 0.012$), and in SIT + RP it increased by 2.9 W ($p = 0.07$). The O₂ consumption increased almost equally, but not significantly, in both groups after treatment by ~ 40 W. The O₂ consumption at baseline was significantly higher for SIT + RP, partly because of a higher metabolic rate indicated by their greater weight (Table I). As a result, the mean MEg remained about the same after treatment in SIT, but increased significantly in SIT + RP by almost 1%. Because of the large variability in baseline values resulting from the differences in motor function in the participants in the two study groups, the three pairs of stair-climbing measurements are shown as percent changes from baseline in Figure 5.

Clearly shown are the significantly greater changes in external work and MEg in SIT + RP than in SIT. The improvement in MEg in the SIT + RP group resulted from a 90% increase in external work that was performed with an increase of only of 23% in O₂ consumed. MEg in the SIT group changed little because their external work and O₂ consumption increased similarly by about 43%. As expected, for all subjects there was a high correlation between baseline values for both external work and MEg with GMFM-66 ($r = 0.79$ and 0.84, respectively ($p < 0.001$ for both). These correlations were also significant within each group ($p < 0.013$). Figure 6 shows that the SIT + RP group increased MEg independently of their baseline GMFM-66, whereas in the SIT group the change in MEg was inversely related to the baseline GMFM-66, this difference between groups being significant.
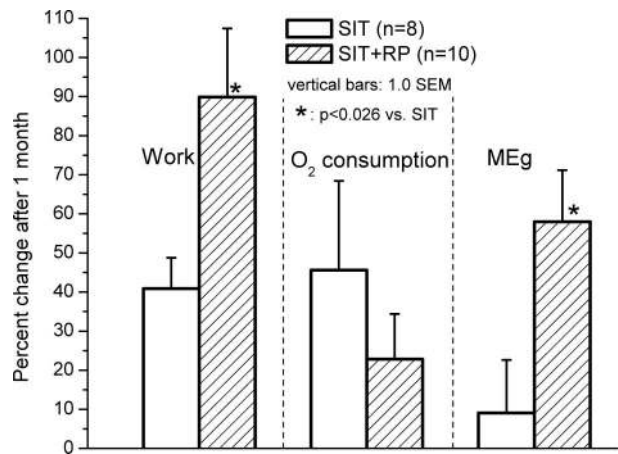

Figure 5. Mean percentage changes for external work (Watts), O₂ cost (Watts) and gross mechanical efficiency (MEg), for study groups before and after treatment. The changes in work and Meg are significantly greater for SIT + RP than for SIT. The effect size, as estimated by Cohen's-d, was 0.51 (medium) for work, 0.06 (low) for O₂ consumption and 0.86 (high) for MEg.
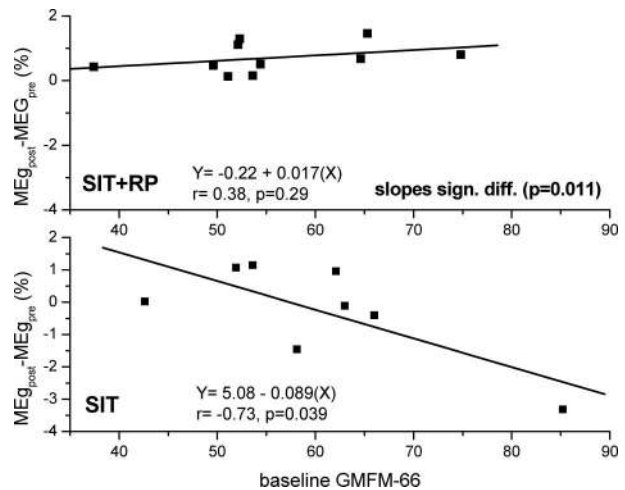
Figure 6. Change in MEg with treatment vs. baseline motor function (GMFM-66). In SIT + RP the changes (increases) are independent of motor function and in SIT they tend to be smaller (negative) in the higher functioning children.

Discussion

This study is an initial attempt to bridge the general theories of dynamic systems with treatment applications. The treatment protocol was designed to determine whether the improvement in gross motor function and mechanical efficiency in children with

CP by intensive therapy would be enhanced by random perturbations that cause a change in motor behaviour. The age range of 6–12 yrs was selected because evidence indicates that children with CP reach about 90% of their gross motor capability at about 5 yrs, depending on GMFCS level [26], and we wanted to evaluate changes in motor functions and efficiency that were not the result of natural development or changes during puberty [27,28].

Therapeutic strategies based on the principle of producing a switch between stability and flexibility of behaviour has been suggested [11,13]. RP was applied using passive random cycling motions for lower and upper extremities, in order to destabilize existing patterns. An important aspect of the coordination of rhythmical movements is the tendency to move the limbs (wings, fins, legs, and arms) at particular frequencies and certain phase relations. This aspect of coordination is called synchronization [29]. We attempted to interfere with these synchronizations and thereby introduce instability to stereotypical activities. We chose cycling as a synchronized activity suitable for children, as the RP intervention. The modified APT was intended to serve in introducing types of RP by applying passive random unsynchronized cycling motions to lower and upper extremities.

When subjects adapt to new external forces, they learn to compensate for these forces by restoring previously learned movements. There is evidence that subjects learn to anticipate external forces when the perturbation does not change [30]. In this study that anticipation was prevented by applying an unexpected external motion, thereby preventing the learning, anticipation and compensation, with the hope of introducing more flexibility. A number of studies have investigated the processes involved in motor adaptation by exposing subjects to specific perturbations and quantifying the changes in their responses over time. These perturbations were usually imposed during the performance of a task in a fixed or random fashion [15,16,31,32]. In this study RP intervention differed from these techniques, in that: (a) RP was applied in passive mode to subjects who were told not to resist it, (b) it was applied before beginning active motor tasks, and (c) test measurements were performed 3–4 days after the intervention program ended and the immediate effects of the RP on the resting muscle tension were no longer present.

RP fits into the theory that the neuromuscular control system must allow for the inherent mechanical properties of inactive muscle fibers to relax in order to control movements with precision and that the motor controller adjusts to changes in resting tension of the muscles [17,33]. Judgments of the importance of clinical changes in relationship to measured changes in GMFM-66 scores are not yet established. For that reason the clinical significance of an improvement of score unit of the GMFM-66 in this study cannot be evaluated. These results suggest that superimposing RP did not contribute to the gain of new motor functions, as measured by this instrument. The GMFM-66 assesses achievement of motor functions, but it is beyond its scope to detect improvement in performance through changes in motor efficiency and control [26]. The greater improvement of MEg in SIT + RP of close to 1% at a self-selected speed may well be physiologically significant. For example, the elite endurance athlete who is now the 7-times winner of the Tour-de France showed an 8% improvement in mechanical efficiency over 7 yrs. Apparently, this improvement in efficiency contributed significantly to a remarkable 18% improvement in his power/kg body weight ratio [34].

Research now suggests that different kinds of interventions can help reduce the energy cost of locomotion. Muscle strengthening exercise programs can improve the patient's ability to compensate for gait disabilities [35]. Aerobic training increases cardiovascular capacity, thus reducing the relative effort of sub-maximal workloads [36]. Both of these modalities can improve the long-term functional capacity of patients with impaired walking ability. Mechanical efficiency was measured here, because of its strong relation to the optimization of motor tasks. There is evidence that humans use energy-minimizing gaits [5]. Models confirm that the force pattern and step frequency that humans adopt at different walking speeds are the ones that minimize the energy cost of locomotion [37].

We hypothesized that enhancing the effects of treatment by introducing flexibility would be manifested by a lowering of the energy cost of ambulation. The stair-climbing test appeared to meet the requirements for the periodic measurement of ambulation in children with CP [38]. The test indicates significantly lower mechanical efficiency in children with CP as compared with ND children [24]. The energy cost of stair-climbing constitutes a comprehensive measure of the components influencing mobility. These components include body and limb mass, range of joint movement and height of step. The MEg measurement is therefore a global and objective assessment of the efficiency of mobility and was chosen to evaluate the efficacy of RP because it incorporates work output of a motor task and is implicit in many definitions of skilled performance.

Improvements in motor function following intensive therapy have been reported [38–40]. This study was designed to address the question of whether children with CP, at the age where they have

plateaued in their motor development, could improve their stair-climbing MEg and whether this improvement is the result of intensive training or gaining more optimal performance of the same task. It has been stated that, 'if more skillful performance decreases the work required to perform a task, motor efficiency has increased,' [22, p. 647] and that mechanical efficiency measurements are sensitive to changes in the coordination and control process of motor functions [41].

The significant improvement in MEg in the study group was the result of a 90% increase in the external work performed at reduced energy cost. The children could perform better with their existing motor abilities and climb more stairs in a given time at their self-selected pace. This improvement was seen in all children in the SIT + RP group, regardless of their baseline motor functions or MEg. In the SIT group the intensive therapy did not to improve the efficiency of children with a higher GMFM-66 and mechanical efficiency at baseline.

Conclusion

This study demonstrates that children with CP were able to adapt to an external random perturbation and improve their mechanical efficiency of stair climbing. The RP apparently weakened their previously established motor behaviour, thus making room for learning to perform more efficient motor tasks. Results suggest that the effect of this kind of intervention is independent of the baseline motor abilities of the child. These results must be confirmed by future studies, but suggest new directions in enhancing therapy outcomes for populations with movement disorders.

Note

1. There are no commercial interests involved.

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