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Motor Control

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Abstract

Background: Cerebral palsy (CP) has been defined as a non-progressive disease of movement and posture development. Physical therapy techniques use different forms of sensory stimulation to improve neuromotor development. **Aim:** The aim of this study was to assess the efficacy of a vestibular stimulation training in improving motor functions in cerebral palsy. **Population:** Fourteen children with CP were randomly separated into two different groups in a cross-over trial. **Methods:** Over a period of 10 weeks, each group performed 10 sessions of 50 minutes of neurodevelopmental treatment (NDT) and 10 sessions of vestibular training (VR). Children were evaluated with the Gross Motor Function Measurement-88 scale, the Goal Attainment Scale and the root mean square of head accelerations. **Results**: A significant improvement in the GAS-score (p=0.003) was noted after NDT+VR. **Conclusions:** Vestibular stimulation integrated with NDT proved to be an effective complementary strategy for facilitating motor functioning.

Key words: vestibular rehabilitation**,** cerebral palsy rehabilitation, neurodevelopmental physical therapy, postural dysfunction

Introduction

Cerebral palsy (CP) has been defined as a non-progressive disease of movement and posture development caused by brain injury during the prenatal, perinatal and postnatal period (Rosenbaum P., 2006). CP is characterized by the inability to control motor functions. It can have an overall negative effect on children’s development because it affects their ability to explore, speak, learn and become independent (Jones MW, Morgan E, Shelton JE, Thorogood C., 2007). Many different physical therapy techniques and approaches are available for treating children with CP. The neurodevelopmental treatments are mainly based on sensory-integration of neural and motor maturation and sensorimotor development (Anttila H et al., 2008; Chrysagis N, Skordilis E, Koutsouki D., 2014; Butler C, Darrah J., 2001; Bobath B., 1964).

A recent study showed that the control of trunk and head orientation and, in turn, navigation is altered in children with CP (Petrarca M. et al., 2013). The Authors suggested the need to develop new therapeutic approaches in which vision is progressively denied to stimulate children to rely on other sensory feedbacks during low and high speed perturbations that require an abrupt change in dynamic coordination.

Among the various techniques using different forms of sensory stimulation to improve neuromotor development, vestibular stimulation could have positive effects on arousal level, visual fixation ability, ocular pursuit movements and motor function in human development (Ottenbacher K., 1983). Ayers in the 1978 suggested the need to carefully consider vestibular stimulation when treating sensory integration because the vestibular system influences all sensory experiences. Premature infants could have sensory deprivation being rarely exposed to abrupt movements and, therefore, receiving little vestibular input (Neal M., 1968). The Authors showed that 30 premature infants receiving vestibular stimulations were more advanced in motor, visual and auditory responses compared to the control group. Several studies confirmed these hypotheses, supporting the idea that controlled vestibular stimulation and multi-sensory stimulations have positive effects on motor development and (Kanagasabai PS. Et al., 2013; Ottenbacher K., 1983) do not just reduce specific vestibular deficits (Ayres AJ., 1978).

A controlled rotatory vestibular stimulation programme produced significant improvements in gross motor skills and in reflex integrations (Clark D, Kreutzberg J, Chee F., 1977). Moreover vestibular inputs through the use of swings may improve the subject's postural control, movement in space, emotional well-being, interaction with people and environment, and participation in play (Sun-Joung Leigh, 2015).

Despite the vestibular stimulations showed positive effects on development of motor control, their integration in a therapeutic approach for children with CP has never been tested. The aim of the present study was to assess the efficacy of vestibular physical therapy, based on the results of previous studies but specifically designed for children with CP to obtain motor function improvement.

Vestibular physical therapy is an exercise-based approach that includes a combination of three different exercise components: exercises to promote gaze stability, 2) exercises to improve balance and gait 3) exercises to facilitate somatosensory integration.

Material and methods

Participants

We carried out a longitudinal, randomized, controlled, cross-over study at the Neuro-Rehabilitation Hospital of Santa Lucia Foundation. From April 2013 to February 2014, all children with a diagnosis of CP admitted to our Hospital were screened by a neurologist for verifying if they matched the above inclusion/exclusion criteria. Among 40 screened subjects, 14 were included into the study.

Demographic and clinical characteristics of the children are reported in Table 1.

Participants were included if they had a diagnosis of cerebral palsy (Jones MW, Morgan E, Shelton JE, Thorogood C., 2007) (mono and bilateral spastic hemiparesis, ataxia), intelligence quotient level (IQ) >49 (i.e. able to understand the tasks), age between of 3 and 11 years, Level of Sitting Scale (LSS) >4 (Field DA1, Roxborough LA., 2012). Exclusion criteria included participation in other experimental studies, conditions in which vestibular rehabilitation is not allowed (such as serious visual fixation disorders), Southern California College of Optometry System (SCCOS) >1(Infants with a score of 1 were excluded because of their changeable fixation), (Hoffman LG, Rouse MW., 1980) severe muscle contractures or bone deformities, skin hypersensitivity (which would prevent application of the accelerometer to the vertex, see below).

After obtainment of approval from the local ethics committee (protocol registration number 380.2) and informed consent from the parents, 14 children were enrolled according to the inclusion criteria and were randomly assigned to one of two arms of a cross-over trial using Research Randomizer Software (Saghaei M., 2011).

Study design

Children were evaluated before the training (T0), at the end of first phase of the trial (T1), at the beginning of the second phase, after a one month wash-out period (T2) and at the end of treatment (T3) as shown in Figure 1.

The assessor physician was blind to the treatment allocations. A wash-out phase was introduced to avoid possible after effects of first training on the second one (so that after effects of the first training could not affect the second one).

During NDT (Aisen ML et al., 2011) each child received ten, 50-minute sessions over a 5-week period. During the 10 sessions of experimental training, each child received 20 minutes of NDT and 30 minutes of VR (which was adapted to our population) over a 5-week period.

NDT and VR were administered by personal physical therapist who had been specifically trained in a post-graduate course. All participants were allowed to use their own orthopaedic shoes or other physical support devices during NDT, but during VR they were not allowed to use walking aids.

Outcome measures

Assessment of the efficacy of the two interventions was performed by a physician using the gross motor function measure-88 (GMFM), the goal attainment scale (GAS) and quantitative tests carried out with a triaxial accelerometer at each assessment-time (T_0, T_1, T_2, T_3), as detailed below (Bjornson KF, Graubert CS, Buford V and McLaughlin J., 1998; Russell DJ. Et al., 1989; Russell DJ. Et al., 2000; Steenbeek D1 et al., 2011). The achievement of each objective was evaluated on a 5 -point Likert scale. Results of the latter allowed creating a profile for each infant; the outcome score and the baseline score were represented by a score of -1. The objectives to achieve with the intervention were selected before the beginning of therapy for each child by the rehabilitation team together with the family. On the GAS, a score of 0 is assigned if the selected objective is reached, +1 if the infant has an improvement little further the expected one, +2 if the improvement is greater than that considered possible, -1 if there are no changes, -2 if the infant worsens. The score obtained is normalized to 100 using the weights defined before treatment when more objectives have been defined. The main objectives were head and trunk stabilization in sitting and standing positions.

The triaxial accelerometer (FreeSense®, Sensorize) was used to provide quantitative data about accelerations along the antero-posterior (AP), medio-lateral (ML) and cranio-caudal (CC) body axes during passive/active locomotion, looking of an object and looking of a video. The triaxial accelerometer is a wearable wireless inertial sensor that is applied over the vertex. The sampling rate was 100 Hz, the range of measurable accelerations ±6g (equal to ±59m/s^2), the weight of the device was less than 100g and data were low-pass filtered at 20 Hz. Several studies have shown the effectiveness of this tool for evaluating postural stability in children (Iosa et al. 2012; Zoccolillo et al. 2015). The device is connected to a computer via Bluetooth. The three components of acceleration, one for each axis, are collected and summarized into their Root Mean Square (RMS) after mean subtraction (Iosa et al. 2012). For the mean subtraction, RMS corresponds to the standard deviation of the signal and provides information about dispersion of the accelerations and hence about instabilities of the head during the three tasks. The accelerometer was fixed over the vertex of the head. Head posture was evaluated: 1) in a sitting position, while the child looked an object at a distance of 2m for 30s (attentiveness task); 2) in a sitting position watching a video at a distance of 1m for 30s (distractor task); 3) during deambulation along a 10m long pathway performed actively (i.e., walking) for children able to perform independent walking, or passively for children confined on a wheelchair that was roughly moved at constant speed (the seatback of wheelchair was removed for assessing the postural control of children). During the looking tasks the children stood up or, when unable, sat in a wheelchair with locked wheels and seatback removed.

Interventions

During the NDT training period participants performed a neurodevelopmental therapy based on Bobath concept (Knox V, Evans AL. 2002), tailored on their individual goals and designed for improving postural alignment, for favoring a more functional muscle tone by specific handling techniques, and for promoting a better motor control for specific and relevant functional skills (Mayston MJ. 2001).

The experimental training was added to NDT and divided into three exercises based on vestibular rehabilitation methods (Han BI, Song HS, Kim JS., 2011), by adapting the methodology to the child’s competencies.

Gaze stability training

Children sat on a chair without the seatback; they were kept in the dark condition to avoid the distraction of any other stimulus and looked at a bright visual target during passive horizontal and vertical head movements (one minute for each axis). Then, if possible, they had to move their head while maintaining their gaze on a firm target (VORx1 one minute for each axis) (Han BI, Song HS, Kim JS., 2011).The therapist checked that the child maintained gaze stability.

Balance and gait training

Children were kept blindfolded and were subjected to changeable stimulations.

This operation consists of two steps with two different exercises to facilitate postural stability in dynamic conditions. For linear accelerations, non-walking children were positioned on a wheelchair without the seatback and subjected to passive transportation in an antero-posterior way. During those movements the children were asked to control their trunk. Intensity and rate of change varied each time, and were adapted to the children's compensatory improvements. Walking children were trained on a treadmill (mean speed: 0.5Km/h) or on carpets with different textures to avoid their being homogeneous.

On average, the exercise lasted 5 minutes for both walking and non walking children.

Angular accelerations were carried out in the same modality for both walking and non-walking children. Infants were positioned on a rotating stool without a seatback. They performed the exercise (each trial lasted about 1 minute) with eyes closed or blindfolded, during passive rotation; the rate varied each time depending on the child's competencies and on their ability to adapt to the stimulation. Both exercises varied progressively in intensity during the sessions to allow the child to be comfortable and stay calm. When the children gained greater knowledge about the exercise and more self-confidence the training became more dynamic and more variable.

Somatosensory integration training

Different systems (i.e.,vestibular, auditory, proprioceptive and motor systems) are involved in this training. It is based on various pertinences and requests, such as recognizing one's own position in space (after active and/or passive angular accelerations), understanding the distance between this position and a target and then looking for it.

Walking and non walking children perform this exercise differently. Non walking children sit on the rotating stool without the seatback. Before beginning the exercise they are positioned in front of a firm target (Voice Output Communication Aid VOCA) that reproduces a chosen auditory stimulation. The blindfolded seated children are subjected to passive angular rotations (both directions), of increasing intensity during the different sessions. During or after rotation they have to reach the target when they believe they are back in the initial position, or they have to look for the target in the space by realizing their position after the rotation. When the children reach the target, they activate the V.O.C.A. and listen to the auditory stimulation chosen. Regarding performance of the exercise by walking children, they perform the angular rotations actively as they stand upright in front of the target.

The firm position of the target was the same as in the first modality. The children actively rotated in both directions while searching for the target.

Statistical Analysis

Results were reported in terms of median and quartiles, together with percentage improvements. Because of the cross-over design of this study, all subjects received both treatment allowing for analyses intra-subject changes in two perfectly matched samples. For NDT+VR baseline scores (T0 for group A and T2 for group B) was compared to the scores recorded at the end of treatment (T1 for group A and T3 for group B). Analogously, for NDT baseline scores (T0 for group B and T2 for group A) were compared to the scores recorded at the end of treatment (T1 for group B and T3 for group A). To analyze also eventual changes during wash-out phases, scores between T1 and T2 were compared within the two groups A and B. The Wilcoxon signed rank test was used to compare the clinical scores (GAS and GMFM) between the baseline and the end of treatment as above described. The percentage changes of acceleration RMS was computed as the difference between the end of treatment and the baseline. Repeated measures analysis of variance (RM-ANOVA) was performed using body axis and group as factors.

The odds ratio, the relevant 95% confidence interval (IC 95%) and the p-value of the chi-squared test were obtained by comparing the number of children who showed an increment (>0) in the GAS-score and the GMFM-score and a reduction in acceleration RMS. The critical alpha level was set at 0.05 for all analyses. The sample size was chosen in accordance with previous similar studies taking into account that the cross-over design increases the power of statistical analyses (Robert MT 2013).

Results

Gender, age and Gross Motor Function Measure were not significantly different between the two groups at T0 (p=0.593, 0.964, 0.128, respectively). The group A that started with NDT+VR adjunction included 4 females and 3 males with a mean age of 7.5±3.4 years and a GMFM score of 60.94±23-17%. The other group included 3 females and 4 males, with age= 7.4±4.6 years and a GMFM score of 58.77±23.17%. One of the 14 enrolled subjects dropped out after the first period of treatment for reasons unrelated to treatment.

GAS

Significant improvement was noted after NDT + VR regarding the GAS-score (median value was 40 at the baseline and 60 at the end of treatment; mean percentage difference was: +42±26%, p=0.003). Conversely, after NDT, the improvement was not statistically significant as shown in Figure 2 (median value at baseline was 45, and it was 50 at the end of treatment; mean percentage differences +9±17%, p=0.063). Wash-out phases did not show any statistically significant change (p=0.317 for group A, p=0.465 for group B).

GMFM-88

Both NDT and NDT + VR induced a significant improvement in the GMFM-score as shown in Figure 3 (mean values, NDT + VR at baseline was 60.9% and it was 63.1% at the end of treatment; the percentage difference was +4±5%, p=0.005; NDT: at baseline was 58.8% and 59.2% at the end of treatment; percentage difference was +1±1%, p=0.034). Wash-out phases did not show any statistically significant change (p=0.102 for group A, p=0.465 for group B).

Accelerations

Distributions of percent acceleration RMS changes were below zero mainly after NDT + VR (median value: -5.6%) not after NDT (median value: + 4.9%). In particular, RM-ANOVA (Table 2) showed a significant interaction between therapy and body axis during forward movement (p=0.044) due to the reduction of ML-accelerations after NDT + VR and the reduction of CC-accelerations after NDT.

Odds Ratios

Odds ratios significantly favourable to NDT + VR were found for the GAS-score (Table 3) and ML stabilization of the head during forward movement and video fixation. Conversely, the odds ratio was significantly favourable to NDT only for CC acceleration during forward movement. No statistically significant odds ratio was found for the GMFM, despite a value of 3.33 that was favourable (but not statistically significant) to NDT + VR with respect to NDT alone.

Discussion

The aim of this study was to assess the efficacy of vestibular rehabilitation training designed specifically for children with CP to obtain motor function improvement. The results obtained with instrumental measures, together with clinical scores support the concept that, compared to NDT, the vestibular rehabilitation training effectively improved both static and dynamic balance. GAS-scores improved significantly only after NDT+ VR, indicating the achievement of the specific objectives this treatment was developed for. Gross motor functions improved after both VR and NDT, because both facilitate physical activity. The measures of stability obtained with the accelerometer revealed much greater improvement overall after NDT+ VR, in particular along the latero-lateral axis. The probability of improving latero-lateral balance (both dynamic and static during video-fixation) was about six times greater after vestibular treatment. However, less improvement of cranio-caudal balance was noted after NDT+ VR with respect to NDT for dynamic balance. This probably means that NDT+ VR improved static balance about six times more than conventional therapy, and with the same proportion modified head instabilities in the frontal plane.

In line with previous results obtained in different populations (Brown KE et al., 2006), our data on vestibular training performed by children with CP resulted in a better stability along medio-lateral axis (as shown by our accelerometric data), and in improving motor control during functional activities (as shown by the results obtained with GMFM and GAS). The fact that NDT was more effective in reducing cranio-caudal instabilities could be related to the use of proper key points or points of control in NDT that facilitated reactions for reacquiring proper posture in terms of vertical stabilization (Bly L., 1991). Nevertheless, in addition to measuring accelerations further studies should investigate this aspect by assessing trunk-neck-head angles to verify possible posture changes. Finally, static balance was improved only in the video-fixation task; this was likely because the children kept their attention on the video during this task; furthermore, fixating an object is more subject to loss of attention. Hence, cognitive factors influenced performances during this task.

For the first time, our results suggest that the processing of sensory stimuli during postural reactions, particularly stimuli from proprioception and from the visual system, are useful and are commonly adopted in traditional therapy. Balance reactions are responses to integrated stimuli that are delivered by proprioceptive and exteroceptive sensations, telereceptors and the vestibular system; therefore, disorders in the reception of these stimuli from any of these receptors or systems can result in impaired stability. Postural reactions are mainly based on vestibular system information, but only the integration of all sensory information affects the final postural response (Kostiukow, A., Rostkowska, E., Samborski, W., 2009).

The present study supports the role of vestibular stimulation training specifically designed for CP children to facilitate static and dynamic balance control. It is conceivable that in these children postural control depends on visual information, whereas in typical development visual orienting should not be essential for maintaining postural stability during quiet standing and walking (Westcott, S. L., Lowes, L. P. Richardson, P. K., 1997).

The vestibular system plays a vital role in ensuring stable body posture as well as gaze. Indeed vestibulo-spinal reflexes play an important role by coordinating head and neck movement with the trunk and body to maintain the head in an upright position. For these reasons we hypotiseze that stimulate the vestibular system through a dynamic training to promote gaze stability and the head/body stabilization could be helpful to facilitate motor control in children with CP.

Moreover facilitate the integration of vestibular information with somatosensory, proprioceptive, and visual inputs as well as motor-related signals also provide selfmotion cues as an organism interacts with its environment.

Similar to many other studies in the field of child neurorehabilitation, the main limits of our study were the small number and the heterogenity of the enrolled children. Nevertheless, the cross-over design and the use of quantitative measures allowed the statistical analyses to have enough power to find statistically significant differences between periods with and without the adjunction of VR. Furthermore, we introduced a wash-out period to avoid that after-effects of the first training affect the second one. On the other hand, the statistical significances we found in our study suggest that it was not underpowered; nevertheless, it should be considered as a pilot study and cautions is needed in result interpretation.

Conclusion

Vestibular physical therapy integrated with NDT could be a dynamic effective complementary strategy for facilitating neuromotor development and stimulating stabilization of head and trunk in both static and dynamic tasks, such as motor control in children with cerebral palsy . In any case, further research is needed to evaluate its long-lasting effects.

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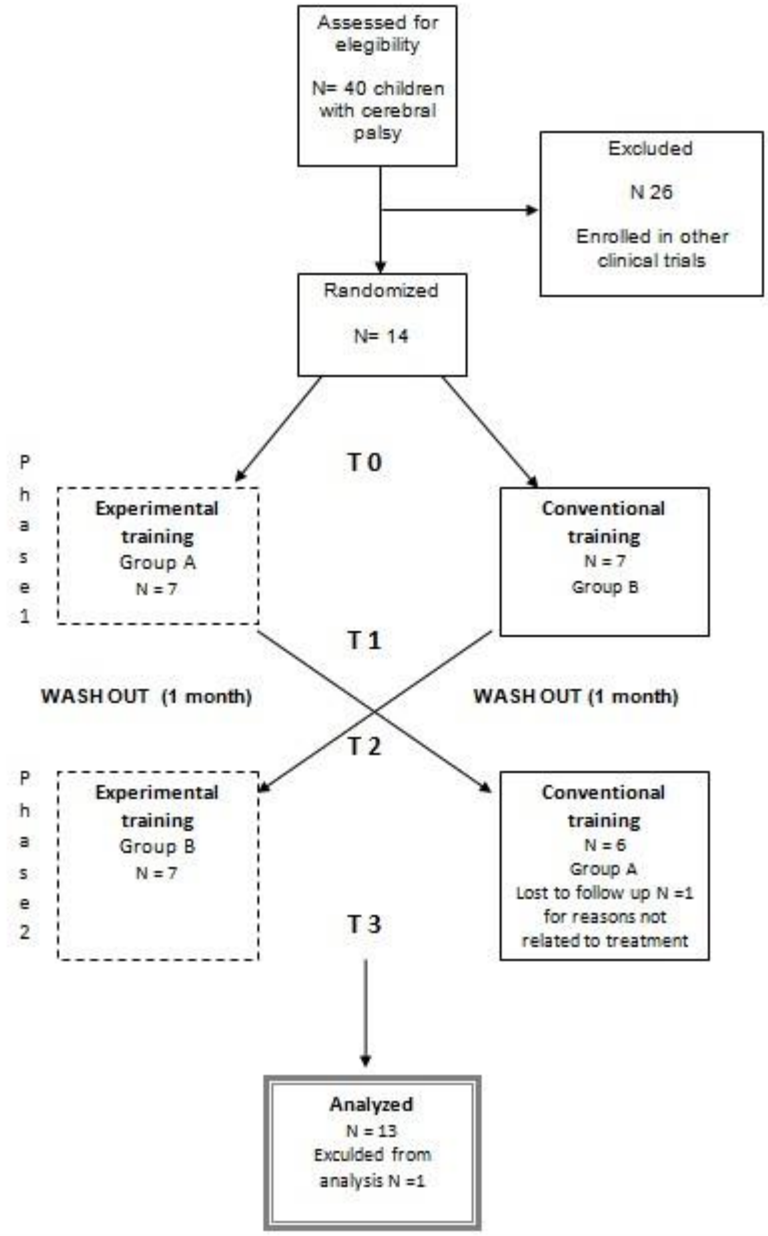

Figure 1. Study design

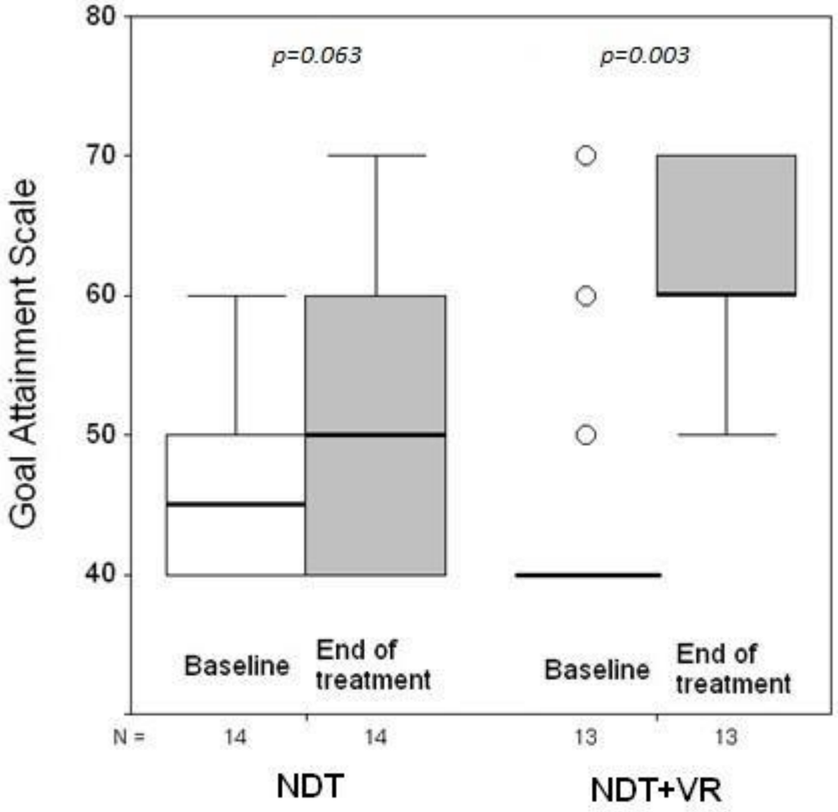

Figure 2. Box-plot of Goal Atteinment Scale score for children during conventional therapy period (left) and experimental therapy period (right). The boxes show the lower quartile, median (bold line), and upper quartile values, the whiskers represent the most extreme values within 1.5 times the interquartile range from the ends of the boxes. Circles represent the values beyond the ends of the whiskers. P-values refers to within subject comparison (Wilcoxon signed rank test).

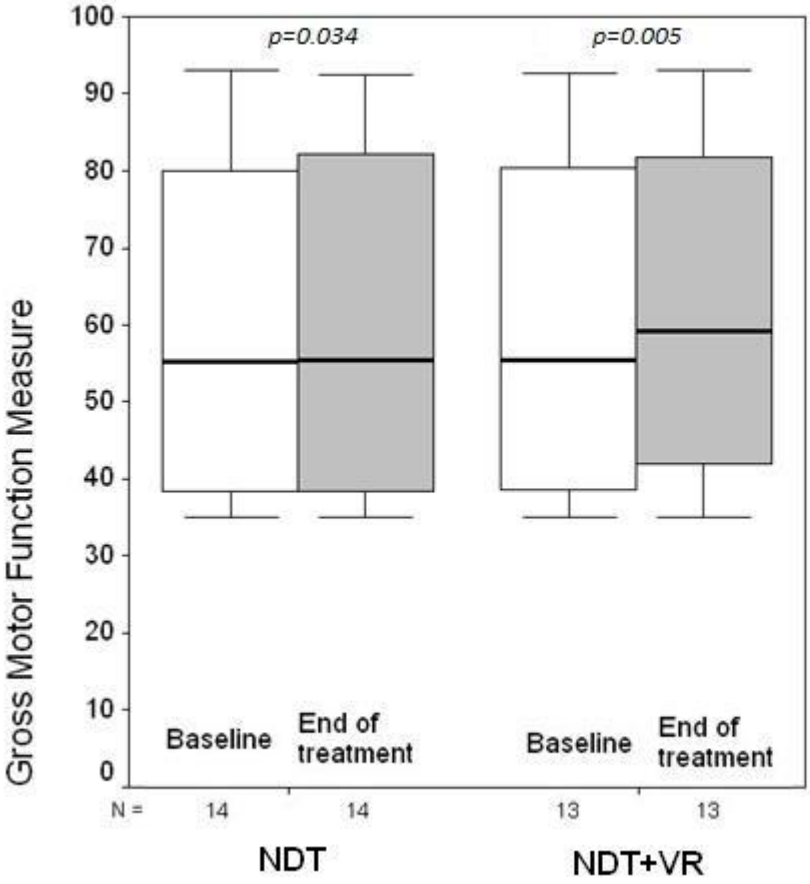

Figure 3. Box-plot of Gross Motor Function Measure score for children during conventional therapy period (left) and experimental therapy period (right). The boxes show the lower quartile, median (bold line), and upper quartile values, the whiskers represent the most extreme values within 1.5 times the interquartile range from the ends of the boxes. P-values refers to within subject comparison (Wilcoxon signed rank test).

***Table 1**. Demographic and clinical characteristics*

Children	***N°***	***Age***	***Gender***	***Mono spastic hemiparesis***	***Bilateral spastic hemiparesis***	***Ataxia***
Group A	7	7.5± 3.4	M = 3 F = 4	2	3	2
Group B	7	7.4± 4.6	M= 4 F= 3	2	3	2

***Table 2**. Results of Repeated Measure Anova*

RM-ANOVA	***GROUP***	***AXIS***	***GROUP*AXIS***
Forward movement	*F(1,12)=0.615* *p=0.448*	*F(2,24)=1.917* *p=0.169*	*F(2,24)=3.580* *p=**0.044***
Object fixation	*F(1,12)=3.261* *p=0.096*	*F(2,24)=2.368* *p=0.115*	*F(2,24)=1.624* *p=0.218*
Video fixation	*F(1,12)=3.140* *p=0.102*	*F(2,24)=0.893* *p=0.423*	*F(2,24)=0.401* *p=0.674*

Table 3. *Odds Ratio of outcome measures results*

Domain	***Outcome***	***OR***	***IC95%***	***p***
Aims	*GAS (>40)*	*9.75*	*0.98-96.56*	***0.029***
Gross motor	*GMFM (+)*	*3.33*	*0.69-16.02*	*0.127*
Forward Movement	*RMS-CC*	*0.16*	*0.03-0.82*	***0.023***
	RMS-ML	*6.60*	*1.23-35.44*	***0.022***
	RMS-AP	*0.49*	*0.09-2.64*	*0.403*
Object fixation	*RMS-CC*	*1.00*	*0.21-4.69*	*0.999*
	RMS-ML	*1.35*	*0.29-6.18*	*0.699*
	RMS-AP	*1.00*	*0.23-4.40*	*0.999*
Video fixation	*RMS-CC*	*1.00*	*0.23-4.40*	*0.999*
	RMS-ML	*6.60*	*1.23-35.44*	***0.022***
	RMS-AP	*1.33*	*0.30-5.91*	*0.705*